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CASE REPORT

Peripartum Cardiomyopathy: A Case Management Series At Sanjiwani Hospital

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Abstract

***Introduction:** In Peripartum Cardiomyopathy (PPCM), heart function will generally return to normal a few months after delivery, but in some cases poor outcomes might occur, therefore early detection and management are crucial. This case series study aims to get an overview of PPCM cases in a type-B referral hospital in Gianyar, Bali.*

***Case Presentation:** This study presents 3 cases of PPCM that showed symptom onset in the antepartum period (the third trimester of pregnancy). Two out of three cases were nulliparous with maternal age <30 years, and went through a Cesarean Section. All three cases underwent treatment in the Intensive Care Unit and showed improvement in their condition.*

***Discussion:** Until now, the heterogeneity of PPCM diagnostic criteria has become an obstacle to its treatment, thus the reported incidence tends to be low. This is likely due to the diagnosis of PPCM which is an exclusion diagnosis from other causes of heart failure. Echocardiography and NT-ProBNP examinations should be done, which, although not specific, may lead to a diagnosis of PPCM when combined with a thorough patient history. The availability of supporting examination modalities in many regions in Indonesia are varied thus referrals are usually needed, resulting in the delay of PPCM cases management.*

***Conclusion:** All pregnant women who experience dyspnea during the third trimester of pregnancy, along with a family history of heart disease, need to undergo close examination and supervision due to the suspicion of PPCM. Early detection and treatment are the main key to successful management of PPCM cases*

***Keywords:** Early Detection; Echocardiography; Peripartum Cardiomyopathy*

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INTRODUCTION

Peripartum Cardiomyopathy (PPCM) is heart failure state caused by left ventricle dysfunction with Left Ventricular Ejection Fraction (LVEF) <45%, occurring at the end of pregnancy or a few months after delivery, with no other identifiable etiology are found[1]. The incidence of PPCM is around 1:2200-4000 (USA), 1:1000 (South Africa), and 1:300 (Haiti). In Asia it is 1:1374 (India), 1:1000 (Japan), 1:837 (Pakistan), 34:100000 (Malaysia)[2]. Studies conducted at Hasan Sadikin Hospital Bandung from January 2011 to December 2013 showed the decreasing incidence of PPCM cases from 2011, 2012 and 2013, namely 51.25%, 27.5% and 21.25%[3].

PPCM has various risk factors, including advanced age, multiparity, obesity, chronic hypertension, severe preeclampsia, eclampsia, and African race. Heart function can return to normal in 23-41% of PPCM patients, thus it is necessary to do an early detection and intervention for reversibility of heart function. Even though the prevalence is low, if it occurs PPCM might cause serious complications thereby increasing maternal mortality and morbidity for the patients[4]. Until now, there has never been a study on PPCM conducted at Sanjiwani Hospital, Gianyar, Bali. This case series study aims to obtain an overview of PPCM cases in a type-B referral hospital in Gianyar, Bali

CASE REPORT

Sanjiwani Hospital is a type-B referral hospital for the east Bali region, with a total of 719 deliveries in 2021. There were 9 cases of mothers with heart disease during the study period, 3 of whom (30%) were diagnosed as PPCM through physical examination and echocardiography. In this study, we report 3 cases of PPCM who came to Sanjiwani Hospital from January 2021 to October 2022.

Case I

A 37 year old pregnant woman, G4P2012 31 week 2 day, came with complaints of intermittent abdominal pain, vaginal discharge, and sudden *dyspnea d'effort* since one day ago. Physical examination showed crackles in both lung and chest wall retraction. The initial electrocardiography (ECG) showed sinus tachycardia, inferior old myocardial infarction, anterolateral ischemia. The patient was diagnosed with G4P2102 31 weeks 2 days, Acute

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Decompensated Heart Failure (ADHF) due to PPCM. Through vaginal delivery, a baby girl was born with a weight of 2300 grams and an APGAR Score of 7-8. The patient was then admitted to the ICU under close monitoring. Echocardiographic examination revealed dilated cardiomyopathy and severe ventricular dysfunction with LVEF of 33.25% and Cardiac Disease in Pregnancy (CARPREG) Score 1. The patient was treated for 5 days (3 days in the ICU and 2 days in the regular ward). The patient was given bromocriptine therapy for 2 weeks along with subcutaneous lovenox anticoagulant and diuretic. The patient's condition has improved with beta-blocker therapy, ACE inhibitors and mineralocorticoid receptor antagonist (MRA) treatment.

Case II

A 27-year-old pregnant woman, G1P0000 37 weeks 6 days, came with complaints of vaginal discharge and sudden orthopnea, since the day before. The patient had a history of hypertension since 37 weeks of gestational age, without complaints of headaches, heartburn, or blurred vision. The blood pressure was 168/108 mmHg. Physical examination revealed no abnormalities of the thorax or extremities. ECG: sinus tachycardia with quadrigeminy ventricular extrasystole (VES). The patient was then diagnosed with G1P0000 37 weeks 6 days, preeclampsia with severe features, premature rupture of membranes, dyspnea, Quadrigeminy VES, Non Reactive NST and planned a Cesarean Section (CS). The patient was then treated in the ICU under close monitoring. A baby girl was born with a weight of 2250 grams and an APGAR score of 8-9. Echocardiographic examination showed dilated cardiomyopathy and severe ventricular dysfunction with LVEF 41.6% - the patient was then diagnosed with PPCM, CARPREG Score 0. The patient was treated for 7 days (5 days in the ICU and 2 days in the regular ward). She received bromocriptine therapy for 1 week, subcutaneous lovenox anticoagulants and diuretics. The patient's condition has improved with beta-blocker therapy, ACE inhibitors and MRA treatment.

Case III

A 22-year-old pregnant woman, G1P0000 35 weeks 4 days, came with complaints of intermittent abdominal pain, sudden paroxysmal nocturnal dyspnea and *dyspnea d'effort*, orthopnea, since 3 days ago. On physical examination, crackles filled both lung and bilateral leg edema was found. ECG revealed sinus tachycardia. The patient was diagnosed with G1P0000 35 weeks 4 days, IUGR, oligohydramnios, sinus tachycardia, Non Reactive NST.

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After the CS procedure, the patient was treated in the ICU under close monitoring. A baby girl was born with a weight of 2100 grams and an APGAR score of 1-3. Echocardiographic examination revealed dilated cardiomyopathy and severe ventricular dysfunction with LVEF of 36.4%, CARPREG Score 3. The patient was given bromocriptine therapy for 2 weeks, subcutaneous lovenox anticoagulants and diuretics. She was treated for 7 days (5 days in the ICU and 2 days in the regular ward). The patient's condition has improved with beta-blocker therapy, angiotensin receptor neprilysin inhibitor (ARNI) and MRA treatment.

Table 1. The Patients' Profile and Clinical Manifestation

	Case I	**Case II**	**Case III**
Maternal Age (years)	37	27	22
Gestational Age	31 weeks 3 days	37 weeks 6 days	35 weeks 4 days
Comorbidity	Hypokalemia	Preeclampsia with severe features	-
GPA status	G4P2102	G1P0000	G1P0000
Delivery method	Vaginal	SC	SC
Neonatal Outcome	Female baby, 2300 gram, APGAR Score 7-8	Female baby, 2250 gram, APGAR Score 8-9	Female baby, 2100 gram, APGAR Score 1-3
Symptoms Onset	31 weeks of gestational age	37 weeks of gestational age	34 weeks of gestational age
Arrhythmia	-	-	Ventricular Fibrillation (VF) /Ventricular Tachycardia (VT)
Maternal Outcome	Improved Condition	Improved Condition	Improved Condition

Tabel 2. Transthoracic Echocardiography Result of The Patients.

	Case I	**Case II**	**Case III**

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LVEF (%)	33.25	41.6	36.4
LVIDs (mm)	50.3	45.1	40.9
LVIDd (mm)	60	56.9	49.6
Pericardial effusion	Absent	Mild	Mild
Mitral insufficiency	Mild	Trivial	Trivial
Tricuspid regurgitation	Mild with high probability of PH (TR Vmax 3.63 m/s)	Absent	Absent
LA (mm)	40	28	23
ECG	Sinus tachycardia, inferior OMI, anterolateral ischemia	VES Sinus tachycardia with VES, Quadrigeminy	Sinus tachycardia

LVEF: Left Ventricular Ejection Fraction; LVIDs: Left Ventricular Internal Diameter End Systole; LVIDs: Left Ventricular Internal Diameter End Diastole; LA: Left Atrium; ECG: Echocardiography

DISCUSSION

Peripartum Cardiomyopathy (PPCM) is a condition of heart failure related to pregnancy, with a causative mechanism that cannot be clearly explained[1,5]. There is a pathogenesis model '*Two-Hit Theory*' which states that the occurrence of PPCM is influenced by 2 conditions: *First Hit* - a hormonal vasculotoxic condition at later gestational age triggers cardiomyopathy; *Second Hit* - there is an underlying genetic predisposition in PPCM (a genome-wide association study (GWAS) found an association between PPCM and a polymorphism located near the PTHLH gene)[6,7]. Diagnostic criterias for PPCM are: 1) Heart failure in previously healthy mothers during the last month to 5 months of postpartum, 2) There is no clear etiology causing heart failure, 3) No documented heart disease before the last month of pregnancy, 4) Evidence of echocardiography results showed reduced LVEF (<45%)[8,9]. Predisposing factors for PPCM are African race, multiparity, multigestational pregnancy, smoking, diabetes mellitus, preeclampsia, advanced age, and family history of cardiovascular

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disease[10,11]. Nevertheless, several studies state that the incidence of PPCM increases in white, primiparous, and younger women[6,7,11,12].

In this study, the three cases showed the onset of sudden dyspnea at the third trimester of pregnancy: at 31, 35, and 37 weeks of gestational age. The symptom onset of PPCM in various studies is uncertain: 75 of 123 patients with PPCM in a case series study in California showed onset of dyspnea during the last month of pregnancy[13] and 35 of 100 cases in the United States complained the symptoms of sudden dyspnea at the first week of postpartum period[14]. Cardiac output increases maximally in the second trimester and tends to plateau in the third trimester, then increases dramatically during labor and postpartum (60-80% above normal), due to increased heart rate and preload from painful uterine contractions and increased catecholamines, as well as autotransfusion of 300-500 ml of blood from uterus to the systemic circulation after each contractions. Cardiac output will continue to increase after delivery due to increased maternal circulation blood volume originating from shifts in uterine and placental blood flow[15].

The clinical manifestations of PPCM may vary depending on the severity of disease at the time of presentation. Symptoms related to heart failure often complained by PPCM patients such as paroxysmal nocturnal dyspnea, leg edema, orthopnea, and *dyspnea d'effort*. Other symptoms include dry cough, palpitations, lightheadedness and chest pain[1,2]. Findings on physical examination such as tachypnea, tachycardia, jugular venous distension, third heart sound, pulmonary rales, and murmurs of mitral regurgitation are common. A small proportion of patients display severe symptoms in the form of cardiogenic shock, severe arrhythmias and thromboembolic complications[10]. In this study, the main symptoms complained by patients in case I was *dyspnea d'effort*; case II was orthopnea; case III was orthopnea, *dyspnea d'effort,* and paroxysmal nocturnal dyspnea. On physical examination, there were crackles in both lung and chest wall retraction in case I; in case II there were no rhonchi or leg edema; and in case III there were crackles in both lung as well as leg edema.

In all the three cases, the initial ECG finding was sinus tachycardia which can be used as a parameter of the patient's prognosis. ECG findings commonly found in PPCM cases are repolarization disorders - QTc interval prolongation and sinus tachycardia can be predictors of a poor prognosis[16]. In case III, there was VT and VF, before finally the patient was referred to our hospital and experienced Return of Spontaneous Circulation (ROSC) after

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cardiopulmonary resuscitation. According to a study by Duncker et al., the incidence of VT/VF in PPCM is associated with a high incidence of sudden cardiac death in young women, as this is related to a complicated genetic phenotype. These ventricular arrhythmias are more common in women with multiparas[17], while the patient in our study is primipara.

The timing and method of delivery in PPCM cases need to involve the patient and be coordinated in a multidisciplinary team. In hemodynamically stable women, delivery can be performed vaginally unless there is an obstetric indication for CS[18] In this study, 2 out of 3 cases underwent emergency CS due to fetal distress, whereas 1 case had vaginal delivery due to stable hemodynamics.

PPCM management according to European Society of Cardiology (ESC) is divided into 5 pillars - BOARD: Bromocriptine (must be followed by anticoagulants due to its thromboembolic feature), Oral heart failure regimens (ARNI, ACE-Inhibitor or ARB and Beta Blocker/Ivabradine), vasorelaxation agents (nitrates/hydralazine) and Diuretics (Furosemide and Spironolactone). The dose and duration of the regimen administration needs to be tailored based on the severity of the PPCM case[19]. In this study, all patients benefited from early ICU care, where close monitoring was performed for acute pulmonary edema, blood pressure, and the risk of developing malignant arrhythmias. There have been no specific recommendations regarding the evaluation of PPCM case monitoring with either laboratory or imaging yet until now. Based on several studies, monitoring is generally carried out by echocardiography at 6 and 12 months after the initial diagnosis. This is related to epidemiological studies where there was significant improvement during the first 6 months[16].

The heterogeneity of diagnostic criteria of PPCM is the most common obstacle found, thus the incidence of PPCM cases reported tends to be low. This is probably due to the diagnosis of PPCM which is the result of exclusion from other causes of heart failure. Echocardiographic and NT Pro-BNP examinations should be done, which, although not specific, can lead to a diagnosis of PPCM when combined with a thorough patient history. The availability of supporting examination modalities in each region, especially remote areas in Indonesia, varies greatly, so that referrals are usually needed, thus the management of PPCM cases might be delayed[16,20]. Moreover, delays in the diagnosis of PPCM also often occur because signs and symptoms of PPCM are generally resemble normal findings in late pregnancy, such as *dyspnea d'effort*, fatigue, orthopnea, paroxysmal nocturnal dyspnea, edema,

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and chest fullness[16]. Another obstacle is the agreement regarding the onset of PPCM symptoms: the initial criteria issued in 2000 by the National Heart, Lung and Blood Institute stated that the onset of PPCM could be from the last month of pregnancy until 5 months postpartum. Nowadays, it was stated that the onset of PPCM has become wider (by ESC guideline) - a few months before birth until one year postpartum[16,21].

In this study, the patients complained of shortness of breath since less than 3 days before hospital admission, thus there was no delay in treatment and led to good outcomes. However, due to the limited number of participants in this case series study, the result of this study cannot be extrapolated to represent the entire population of people in the Bali Province. Although cardiac function can return to normal in 23-41% of PPCM patients[4], some cases experience poor outcome[22], therefore early detection through risk factors investigations, as well as early treatment and close monitoring is the main key to managing PPCM cases.

CONCLUSION

All pregnant women who complain shortness of breath (orthopnea, paroxysmal nocturnal dyspnea, *dyspnea d'effort*) during the third trimester of pregnancy, accompanied by a family history of heart disease, need to undergo close examination and supervision because of suspicion of PPCM. Early detection and treatment is the main key to successful management of PPCM cases

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