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CASE REPORT

A Very Rare Cause of Cardiomyopathy in a Child: Peripartum Cardiomyopathy

Çocukta Çok Nadir Bir Kardiyomiyopati: Peripartum Kardiyomiyopati

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ABSTRACT

Aim: Peripartum cardiomyopathy (PCMP) is characterized by severe systolic dysfunction of the heart, manifesting with signs and symptoms of heart failure in the last month of pregnancy or within the first five months postpartum in women with no previously known heart disease.
Methods: An adolescent mother presented to our emergency department at three months postpartum with complaints of dyspnea, orthopnea, edema, and palpitations.
Results: Following the diagnosis of PCMP, standard cardiomyopathy treatment was initiated alongside bromocriptine therapy. However, during follow-up, the patient died due to heart failure.
Conclusions: PCMP, which is rare even in adult patients, can also occur in the pediatric age group. The use of bromocriptine, which pediatric cardiologists may be less familiar with, in addition to standard heart failure therapy, may be required for effective treatment, but the nature of the disease could be very severe.

Keywords: Adolescent, bromocriptine, cardiomyopathy, peripartum, pregnancy

ÖZ

Amaç: Peripartum kardiyomiyopati (PCMP), öncesi bilinen bir kalp hastalığı olmayan kadında hamileliğin son ayında veya postpartum ilk beş ayı içinde kalp yetersizliğinin işaret ve bulguları ile beraber kalbin ağır sistolik disfonksiyonu ile karakterizedir.
Gereç ve Yöntemler: Doğum sonrası 3. ayında dispne, ortopne, ödem ve çarpıntı şikayetleri ile acilimize adolesan anne başvurdu.
Bulgular: PCMP tanısı konularak, standart kardiyomiyopati tedasine ek olarak bromokriptin başlandı. Bununla beraber izlemde hasta inatçı kalp yetmezliğinden kaybedildi.
Sonuçlar: PCMP, erişkin hastalarda dahi nadiren gözlenir, çocukluk yaş grubunda da oluşabilir. Çocuk kardiyologlarının daha az aşina olduğu, bromokriptine kullanımı, standart kalp yetersizliği tedavisine ek olarak, hastalığı iyileştirmek için gereklidir, ancak hastalığın doğası çok ağır olabilir.

Anahtar Kelimeler: Adolesan, bromokriptin, gebelik, kardiyomiyopati, peripartum

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INTRODUCTION

Peripartum cardiomyopathy (PCMP) is a rare disease that causes systolic dysfunction of the heart in women without previously diagnosed heart disease, usually established through specific diagnostic criteria and by excluding other potential causes of heart failure. The Demakis criteria, defined by Demakis et al. (1,2), are as follows: Heart failure developing in the last month of pregnancy or within the first five months postpartum, no previously identified heart disease, exclusion of other causes of heart failure, and echocardiographic findings such as left ventricular dysfunction (ejection fraction <45%). The poor prognosis of PCMP developing in an adolescent pregnant woman should be kept in mind, as this disease can also be seen in childhood.

Case Report

A 14-year-old female patient, a Syrian refugee, presented with a 3-month history of easy fatigability, respiratory distress, palpitations, and particularly severe nocturnal dyspnea. She came to our pediatric emergency unit complaining of diffuse edema, weakness, and palpitations. A more in-depth history revealed that she had become pregnant at age 13 and was not followed by an obstetrics specialist during pregnancy. She reported no prior cardiac or other complaints. There was no remarkable family history. Apart from partial albinism, there were no notable features in her personal medical history (Figure 1).

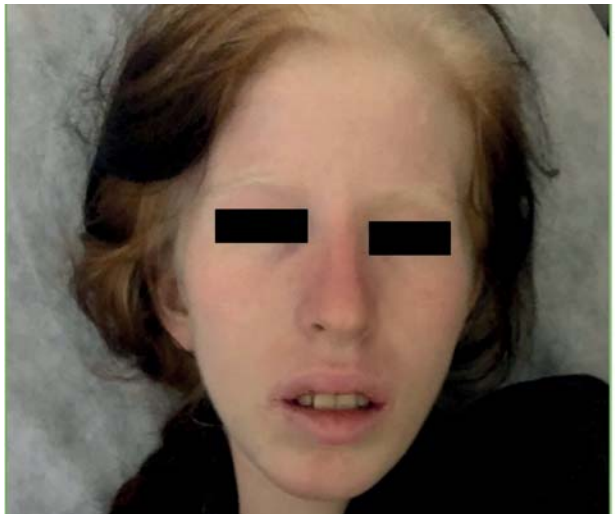

Figure 1. The patient with partial albinism and peripartum cardiomyopathy

Cardiac auscultation revealed gallop rhythm and a grade 3/6 murmur. She had tachypnea, dyspnea, and orthopnea; crepitant rales were noted, especially at both lung bases. Her abdomen was distended with 3 cm of hepatomegaly. On chest radiography, the cardiothoracic ratio was increased. An electrocardiogram showed ST depression in the V6 lead (Figure 2). Transthoracic echocardiography of the left ventricle showed: Left ventricular end-diastolic diameter 6.8 cm, end-systolic diameter 6.1 cm, ejection fraction 20%, fractional shortening 9% (Figure 3). Mitral regurgitation of grade 3 secondary to annular dilation was present.

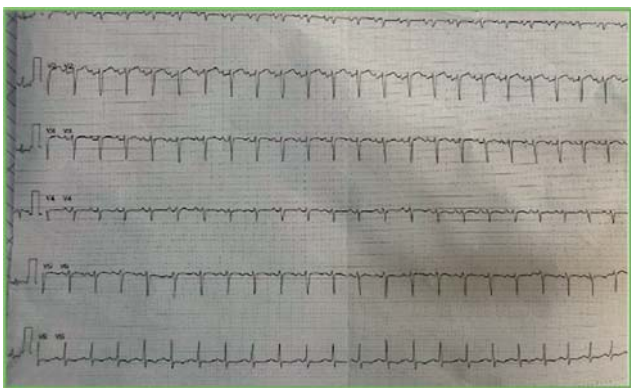

Figure 2. Electrocardiogram showing T-wave inversion in lead V6.

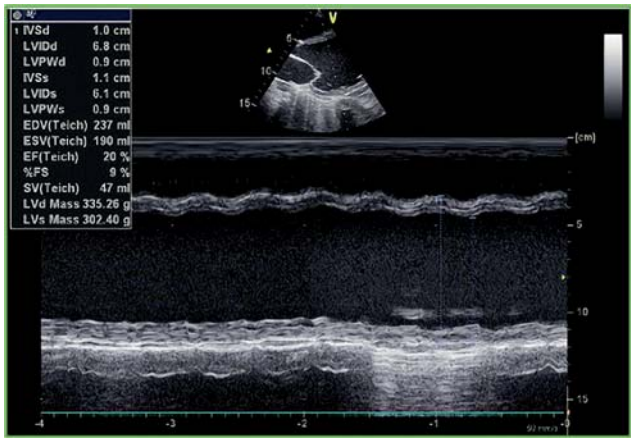

Figure 3. M-mode echocardiogram demonstrating left ventricular dysfunction.

Her brain natriuretic peptide level was very high, 3094 ng/L. Infection markers, blood gas analyses, troponin values, and other blood tests were within normal limits. Viral serology and metabolic screening were unremarkable. Abdominal ultrasonography showed widespread free fluid in the lower abdominal and pelvic regions. A brief run of ventricular tachycardia was detected on Holter recording (Figure 4).

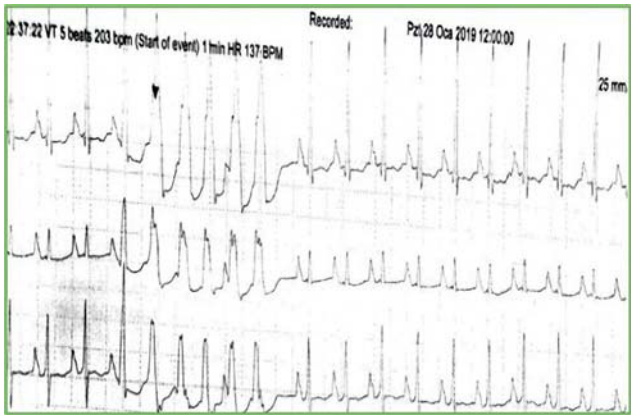

Figure 4. A Holter recording capturing a brief 5-beat run of ventricular tachycardia.

According to the New York Heart Association functional classification, the patient was in class IV and met all Demakis (1,2) criteria for peripartum cardiomyopathy (PCMP); therefore, the case was admitted with the diagnosis of postpartum cardiomyopathy. Intravenous diuretics, dopamine, dobutamine, and oral angiotensin-converting enzyme inhibitor therapy were started. Nasal oxygen support was given. During follow-up, the patient remained hypotensive with increasing oxygen requirements; maximal doses of inotropes were administered, and milrinone was added. Considering the possibility of viral myocarditis, intravenous immunoglobulin therapy was given. On the 4th day of admission, bromocriptine was added to the existing therapy, and breastfeeding was discontinued. Bisoprolol was added as antiarrhythmic therapy. On the 14th day of hospitalization, her general condition suddenly deteriorated, and she was transferred to the intensive care unit with preparations for possible heart transplantation. Unfortunately, the patient died on the 14th day of hospitalization due to heart failure. Informed consent was obtained from the patient's parents to publish the medical condition.

DISCUSSION

Our patient fulfilled all these criteria and was diagnosed with PCMP. Although the true incidence is unknown, regional differences exist, likely due to environmental and genetic factors, cultural practices, and variations in perinatal care (2). The ARTEMIS study conducted in Turkiye evaluated 293 adult women diagnosed with PCMP, revealing a relatively high all-cause mortality rate of 5.1%, while bromocriptine was administered in only 6.9% of cases (3).

In PCMP, overexpression of the 16 kDa prolactin fragment has been implicated. This fragment is thought to trigger and maintain the oxidative stress that results in myocardial dysfunction and heart failure (2). Prolactin is considered central to the cascade of events leading to oxidative stress in postpartum cardiomyopathy (2,4). Bauersachs et al. (1) identified the cleavage of prolactin into its cardiotoxic fragment under oxidative stress as a major driver in PCMP pathophysiology. Prolactin suppression accelerates recovery and improves systolic function. Patients treated with bromocriptine demonstrated greater improvement in left ventricular systolic function than those who did not receive the drug, with a more sustained improvement over a 6-month follow-up (2,5).

In our patient, after five days of bromocriptine, an improvement in systolic function and a notable reduction in BNP levels were observed. Despite this, the patient's heart failure progressively worsened, and she passed away on the 14th day of hospitalization. PCMP is a condition where early diagnosis and treatment are critical. Delayed diagnosis (beyond 3 months postpartum) and the late initiation of bromocriptine may have negatively impacted our patient's clinical course. We thus believe that adding bromocriptine early in the course of disease to standard therapy for cardiomyopathy could be beneficial. Bromocriptine is a semi-synthetic ergot alkaloid and a dopamine agonist. It is metabolized by the cytochrome P450 system and excreted by the liver. Due to its ability to block prolactin release from the pituitary gland, it has long been used in the treatment of hyperprolactinemia (6).

According to the EURObservational Research Programme Peripartum Cardiomyopathy (EORP PPCM) registry published in the European Heart Journal in 2023, patients treated with bromocriptine had a lower risk of adverse maternal outcomes, such as death, rehospitalization, or left ventricular ejection fraction (LVEF) below 35%, during a 6-month follow-up. In this study, the rate of adverse outcomes was 22% in patients treated with bromocriptine, compared with 33% in those treated with standard therapy ($p=0.044$) (6). In addition, multivariate analyses showed that bromocriptine treatment reduced the risk of adverse maternal outcomes (OR: 0.47; 95% CI: 0.31–0.70; $p<0.001$) (7).

Despite major advances in the diagnosis and treatment of the disease, many women still die from it or suffer from permanent cardiac problems. For this reason, care should be taken in very early age pregnancies as well as in older ages. Early motherhood presents high risks for both maternal and neonatal outcomes; adolescents are often physically unprepared for childbirth, leading to higher rates of complications and mortality. The actual frequency of early marriages is challenging to ascertain due to underreporting of unofficial marriages and difficulties with data collection methods (8).

According to the Turkish Statistical Institute, the rate of marriages under 18 was 3.5% in 2022 and 3.1% in 2023 (9). These figures include only official marriages, excluding undocumented marriages and religious unions. Among refugees in Turkiye, early marriages are often unregistered and performed by religious ceremonies, complicating the data further. Although advanced maternal age is a known risk factor for PCMP, there are no published

pediatric cases of PCMP in the literature despite extensive searches.

CONCLUSION

As child marriages among refugees increase, pediatric cardiologists may encounter PCMP in younger age groups. We propose that both extremes of maternal age—very advanced and very young—may increase the risk for this condition.

Conflict of interest: None

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