

Neonatal infective endocarditis in a cardiac rhabdomyoma case: A rare presentation

Manifa Afrin¹, Anima Ferdous², Monir Hossain³

From ¹Associate Professor, Department of Pediatrics, Universal Medical College, ²Specialist, ³Chief Consultant, NICU and PICU, Universal Medical College Hospital, Dhaka, Bangladesh

ABSTRACT

Neonatal infective endocarditis (IE) is an exceedingly rare disease and usually not associated with cardiac rhabdomyomas or any underlying structural cardiac anomalies. Cardiac rhabdomyoma is also the most common benign primary cardiac tumor. The prognosis depends on the size, location of tumors, and outflow tract obstruction but can regress within 2 months of age and reduces the necessity of surgery. Due to the variable clinical features and course, we need to evaluate cardiac vegetation as soon as possible for better outcomes. A combination of these two conditions was not reported before. Here, we presented a case of IE with cardiac rhabdomyoma in a male baby which is first reported from Bangladesh previously.

Key words: Cardiac rhabdomyoma, Cardiac, Neonatal infective endocarditis, Tumor

Primary cardiac tumor is a rare entity with only 0.027% incidence in children [1-3]. Cardiac rhabdomyoma is the most common benign tumor in a fetus with an incidence of 60–86% and in infants, the incidence is 45% as a primary tumor [1]. About 50% of cardiac rhabdomyoma are associated with tuberous sclerosis and about 100 cases have been reported in different literature [4,5]. Neonatal infective endocarditis (IE) is also a rare disease but without any underlying structural heart anomalies [6].

Here, we reported a case of IE with cardiac rhabdomyoma with an absence of tuberous sclerosis which was an unusual presentation. As far as our knowledge, the combination of these diseases is first reported from Bangladesh previously.

CASE REPORT


A male baby was born to a 24-year-old 3rd gravid 2nd Para mother at 35 weeks of gestation, by cesarean section due to scar tenderness, gestational diabetes mellitus (on insulin), and previous cesarean section. At birth, he needed resuscitation as stimulation, nasopharyngeal-oral suction, and oxygen inhalation with an Apgar score of 6/10. Soon after birth, the baby developed respiratory distress and need oxygen through a face mask and head box. But the baby was not improved and put in

continuous positive airway pressure (CPAP) care for 9 days. His condition again deteriorated and needed mechanical ventilation (MV) from 11 days of his age.

At 18 days of age, the baby was severely dyspnoic, cyanosed, in shock, edematous, pale, and bleeding from the nose. The baby was resuscitated with two boluses of normal saline, dopamine (10 µg/kg/min), dobutamine (10 µg/kg/min), and continued MV care with injection ceftazidime and amikacin. The baby's condition became worsen despite all supportive measures.

His investigation revealed hemoglobin 12.6 g/dl, the total count of white blood cell 13,000/cmm, neutrophil 74%, lymphocyte count 20%, platelet 80,000/cmm, hypoalbuminemia, hypokalemia, positive C-reactive protein (CRP), and prolonged prothrombin time. The chest X-ray revealed bilateral pneumonia and an echocardiogram described large vegetation (8.7 mm×2.8 mm) attached to anterolateral papillary muscle with small vegetation (4.3 mm×2.5 mm) attached to the posterior mitral leaflet (PML). There was also moderate persistent pulmonary hypertension (PPHN) with pulmonary artery systolic pressure (PASP) of 46 mmHg, good left ventricular function, ejection fraction (EF) of 61% as shown in initial echocardiography (Fig. 1). The culture report shows that *Candida* species and *Escherichia coli* were sensitive to imipenem, gentamicin, and fluconazole.

Since the baby was pale (Hb 9.7 gm/dl) with thrombocytopenia (36,000/cmm), and increased CRP, MV care was provided to the baby along with transfusion of blood and blood products. Albumin

Access this article online	
Received - 18 June 2021 Initial Review - 03 July 2021 Accepted - 31 July 2021	Quick Response code 
DOI: 10.32677/IJCR.2021.v07.i08.2986	

Correspondence to: Dr. Manifa Afrin, Department of Pediatrics, Universal Medical College, 74G/75, Peacock Square, New Airport Road, Mohakhali, Dhaka 1215, Bangladesh. E-mail: manifaaftrin@gmail.com

© 2021 Creative Commons Attribution-NonCommercial 4.0 International License (CC BY-NC-ND 4.0).

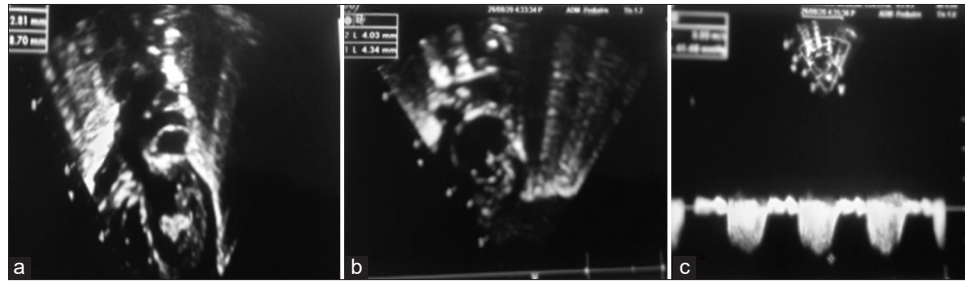


Figure 1: (a-c) Large vegetation (8.7 mm×2.8 mm) attached to anterolateral papillary muscle

infusion was given repeatedly. In addition to that, antibiotics were given to the baby. On the 23rd day of life, the patient was slightly stable and was weaned to CPAP care. However, while on CPAP and desaturation, the patient again developed respiratory distress. The baby was reintubated and kept on MV care with a high set up for the next 3 days. Again the baby became stable within 5 days with lower ventilatory parameters.

Due to slow improvements, a repeat echocardiography was done which had shown a large cardiac rhabdomyoma (22 mm×8 mm) attached to the anterolateral papillary muscle of the left ventricle obstructing the outflow tract along with IE of mitral valve (5 mm×3.4 mm) attached to PML and mild pulmonary arterial pressure (PASP: 30 mm of Hg) with good biventricular function (EF: 62) (Fig. 2).

The patient was again weaned to CPAP care, then subsequently, CPAP was withdrawn and kept on head box to face mask. The baby showed gradual improvement after 1 month of age, maintaining oxygen saturation in room air and was transferred to the ward from NICU at 33 days of age.

DISCUSSION

Neonatal IE is usually a rare and fatal condition [7], previously not diagnosed because of unavailability of diagnostic tools like an echocardiogram. Nowadays, awareness and early detection reduce the fatality rate [8]. Congenital heart disease is found in only 8% of neonates as compared to 80% in older children and adults with IE [6,9,10].

Risk factors for neonatal IE are invasive umbilical and other central lines, total parenteral nutrition, and septicemia. For non-bacterial thrombotic endocarditis, important risk factors are hypoxia in a neonate due to perinatal stress, low APGAR score at birth, low pH at birth, resuscitated neonate, PPHN, hyaline membrane disease. Positive blood culture, respiratory distress, new murmur, hematuria, congestive cardiac failure (CCF), fever, and central lines are seen in both types of IE [6,11]. Our patient has septicemia, persistent thrombocytopenia, required resuscitation after birth, and was culture-positive with echocardiographic evidence. This presentation is similar to Afif's findings [12]. Clinical presentations are non-specific, variable and depend on the highest suspicions of clinicians [8].

There are no definite diagnostic criteria for neonates but still, echocardiography and blood culture are usual diagnostic tools. Thrombocytopenia is evidence of septicemia and taking longer

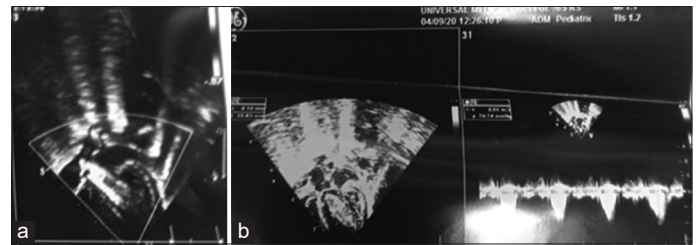


Figure 2: (a&b) Large cardiac rhabdomyoma (22 mm×8 mm) attached to anterolateral papillary muscle

than usual time to resolve [6]. Still, the mortality rate is 25–60% at present, it can be possibly decreased by early diagnosis and treatment with higher dose antibiotics than for sepsis [13]. Fungal infections are rare, no case report was found as before. In our baby, *Candida* species were found in the blood.

Cardiac rhabdomyoma is an extremely rare childhood tumor [14]. Infant cardiac tumors are determined by postmortem which are not possible in resource-limited areas [15]. The tumor has different clinical presentations and surgery was done in hemodynamically unstable conditions [2]. Clinically undetectable but discovered during investigation for other conditions such as heart failure, arrhythmia, or fetal ultrasound for any purpose [16]. Important features are spontaneous regression but may cause serious clinical manifestations before regression such as severe arrhythmias, ventricular inflow and outflow tract obstruction, CCF, and even death. These patients need medical care until the condition is not suitable for intervention [2]. Our patient had a large mass and evidence of IE, where surgical correction is not possible for general wellbeing.

As the maternal transplacental transmitted estrogen is responsible for rhabdomyomas growth, regression occurs with the first 2 months of life due to reduction of this effect [3]. Asymptomatic patients need close monitoring which can assess prognosis and regression status with exclusion of association as tuberous sclerosis [3,11,14,17]. In tuberous sclerosis, stigmata presents in families [17] but our baby had no such history.

Surgical intervention is not necessary, depending on extend of outflow obstruction and failure of regression. Because of changing and unstable nature of this tumor, the clinical presentation is not constant and also prognosis is varied except association of cardiac failure. According to Durairaj *et al.*, 85% of cases had documented spontaneous regression with a good prognosis [15].

Individual diseases are reported before from different countries and institutions [3,6,11,13,14]. Due to atypical and uncommon

disease of cardiac rhabdomyoma with IE in neonates, very few reports are presented in the literature. Hence, we have tried to present the details of this type of rare case from a resource-limited country like Bangladesh.

CONCLUSION

We report a case of cardiac rhabdomyoma located in the left anterolateral papillary muscle of the left ventricle which obstructs the outflow tract and IE of the mitral valve which was improved after conservative treatment. With improved sonographic technology, the diagnosis of cardiac rhabdomyomas and IE may become easier, earlier, and more accurate in the clinical diagnosis and treatment era.

AUTHOR CONTRIBUTION

Conception and design, acquisition, interpretation, drafted the manuscript, revised the manuscript.

REFERENCES

1. Chang J, Chiou P, Yao S, Ching Chou I, Lin CL. Regression of neonatal cardiac rhabdomyoma in two months through low-dose everolimus therapy: A report of three cases. *Pediatr Cardiol* 2017;38:1478-84.
2. Bornaun H, Oztarhan K, Erenner-Erhan T, Dedeoğlu R, Tugcu D, Aydoğmuş C, *et al.* Regression of cardiac rhabdomyomas in a neonate after everolimus treatment. *Case Rep Pediatr* 2016;2016:8712962.
3. Tzani A, Doulamis IP, Mylonas KS, Avgerinos DV, Nasioudi D. Cardiac tumors in pediatric patients: A systematic review. *World J Pediatr Congenit Heart Surg* 2017;8:624-32.
4. Marx GR, Moran MA. Cardiac tumors. In: Allen HD, Gutgesell HP, Clark EB, editors. *Moss and Adams' Heart Disease in Infant, Children and*

Adolescents. 6th ed. Philadelphia, PA: Lippincott Williams and Wilkins; 2000. p. 1435-7.

5. Farooki ZQ, Ross RD, Paridon SM, Humes RA, Karpawich PP, Pinsky WW. Spontaneous regression of cardiac rhabdomyoma. *Am J Cardiol* 1991;67:897-9.
6. Dhungana SP, Shrestha S, Shrestha SK. Infective endocarditis in a neonate. *J Nepal Paediatr Soc* 2017;37:286-9.
7. Soo SS, Bosman DL. *Streptococcus fecalis* in neonatal infective endocarditis. *J Infect* 1991;23:209-10.
8. Nadroo AM, Wear AA, Sowailam AM, Jubair FA. Infective endocarditis in a neonate. *Ann Saudi Med* 1997;17:347-9.
9. Callaghan O, McDougallie P. Infective endocarditis in neonates. *Arch Dis Child* 1988;63:53-7.
10. Millard DD, Shulman ST. The changing spectrum of neonatal endocarditis. *Clin Perinatol* 1988;15:587-607.
11. Uzun O, Wilson DG, Vujanic GM, Parsons JM, De Giovanni JV. Cardiac tumors in children. *Orphanet J Rare Dis* 2007;2:11.
12. El-Khuffash A, Molloy EJ. Incidental finding of endocarditis in a preterm neonate. *Arch Dis Childhood* 2007;92:F134.
13. Mecrow IK, Ladusans EJ. Infective endocarditis in newborn infants with structurally normal heart. *Acta Paediatr* 1994;83:35-9.
14. Bassirou MB, Mohamed L, Fatou A, Alassane AN, Abdoulaye C, El hadj Yaya B, *et al.* Infant cardiac rhabdomyoma: About a case. *J Clin Exp Cardiol* 2020;11:657.
15. Durairaj M, Mangotra K, Makhale CN, Shinde R, Mehta AC, Sathe AS. Cardiac rhabdomyoma in a neonate: Application of serial echocardiography. *Echocardiography* 2006;23:510-2.
16. Gournay V. Children's heart tumors. *Pediatr Realit* 2014;184:24-8.
17. Aggarwal M, Sachan R, Arya S, Chellani H. Cardiac rhabdomyoma--a case report. *J Trop Pediatr* 2010;56:345-7.

Funding: None; Conflicts of Interest: None Stated.

How to cite this article: Afrin M, Ferdous A, Hossain M. Neonatal infective endocarditis in a cardiac rhabdomyoma case: A rare presentation. *Indian J Case Reports*. 2021;7(8):357-359.