

**Title: A rare entity of Double Chambered Right Ventricle with Ventricular Septal defect, a congenital heart disease: A case report**

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**Abstract:**

**Introduction:**

The double or dual-chambered right ventricle is a rare congenital heart defect having two separate pressure compartments in the right ventricle (RV) and is frequently accompanied by Ventricular Septal Defect (VSD). An atypical bundle of muscle crossing the RV from the interventricular septum to the RV-free wall usually causes the blockage. Hypertrophy of the oblique component of the bulbar muscle causes the blockage, which is located at the commencement of the outflow tract next to the moderator band. The ventricular septal defect is frequently obscured due to beginnings of muscle bundles from the upper half of the interventricular septum. This case is of a 6-year-old boy with left-handed dominance residing in a rural area. As narrated by the mother, about one month back he started feeling dyspneic during playing, crying, and defecating (Grade 3 on the NYHA scale) along with diaphoresis while exertion like running and prolonged playing.

**Conclusion:**

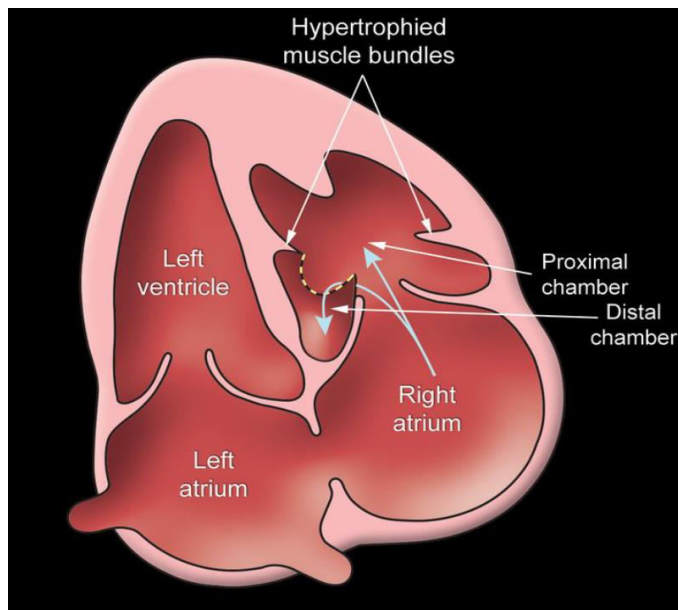
Cardiovascular and pulmonary physiotherapy is an evidence-based practice that evolved rapidly in a few decades and works parallel to the medical and surgical team's intervention. It is a cost-effective form of treatment that rapidly helps in the betterment of patients suffering from pulmonary as well as cardiac illnesses in in-patient & out-patient departments as well as Intensive Care units.

**Keywords:** Double chambered right ventricle, muscular septum, Ventricular septal defect, congenital heart disease, Physiotherapy.

**Introduction:**

DCRV (dual-chambered right ventricle) was initially reported in late 1850s by TB Peacock, but it is today considered as a kind of congenital cardiac disease comprising of the right ventricle's division into a proximal component of high-pressure and a distal portion of low-pressure due to a mid-cavitary blockage. DCRV is divided into two categories based on the tissue that causes the mid-cavitary blockage. Type 1 DCRV patients have an abnormal muscular bundle that spans the right ventricle, whereas type 2 DCRV patients have parietal and septal muscle hypertrophy that causes a blockage, and they are more likely to have a ventricular septal defect (VSD) (Patibandla and Kyaw, 2022). VSD (the most frequent and seen in more than 83% of DCRV cases), transposition of the great arteries, tetralogy of Fallot, ruptured sinus of Valsalva aneurysm, atrial septal defect, aortic valve regurgitation, persistent left superior vena cava, tricuspid valve regurgitation, Ebstein anomaly, and valvar pulmonary stenosis are all associated abnormalities (Gurbuz *et al.*, 2015). The formation of aberrant bundles of muscle in patients with double-chambered right ventricle is explained by a variety of causes, according to theories. The following are some of them:

1. Hypertrophy of the supraventricular crest results from increased blood flow in individuals with VSD, culminating in acquired blockage.
2. The septomarginal trabecula, also known as the moderator band, experiences gradual hypertrophy over time due to superior displacement.
3. The establishment of obstructive muscle bundles is caused by the extension of a muscular shelf from the septoparietal trabeculations to the trabecular component of the right ventricular apex in combination with other acquired variables(Darwazah *et al.*, 2011).



The embryological foundation for a double-chambered right ventricle is considered either a failure to integrate the bulbus cordis into the RV or a raised hypertrophied moderator band. Byrum *et al.*, on the other hand, examined the pattern of electrical activity to conclude that muscle bundles were not the product of a misplaced moderator band and proposed that activation of the double-chambered right ventricle is identical to that of the normal heart(Byrum *et al.*, 1982).

### Patient Information:

This case is of a 6-year-old boy with left-handed dominance residing in a rural area. As narrated by the mother, about one month back he started feeling dyspneic during playing, crying, and defecating (Grade 3 on the NYHA scale). He also experienced profuse sweating while exertion like running and prolonged playing. The mother additionally reported a history of complaints of increased work of breathing and respiratory distress at the age of 2 months of the

patient. When visited by a local practitioner, his results divulged that he was diagnosed with congenital heart disease due to severe supra-pulmonary stenosis. Symptomatic treatment was provided which was helpful and improved his symptoms in 2-3 weeks. In due course, his milestones were achieved at the proper age as per his parents. However, the parents were counseled about the severity of the disease and advised a surgical correction for the same at the age of 5 years. On admission to our tertiary care hospital, on 19/01/2022; a repeat 2D Echocardiography was done which showed a Double Chambered Right Ventricle along with a Ventricular septal defect. Surgical intervention was planned by cardiothoracic surgeons and the patient was referred to Cardiovascular and pulmonary physiotherapists for pre-operative physiotherapy on 21/01/2022. Surgical correction was Intra-cardiac Repair (ICR) done on 25/01/2022. The patient was kept in ICU for 3 days under observation and then shifted to the CVTS ward on 28/01/2022, while his Physiotherapy management was continued.

### Clinical Findings:

He was assessed in supine lying position with head end of his bed elevated to 30 degrees. His neck was in slightly flexed, shoulders protracted, elbows and wrists in extension bilaterally; his bilateral hip, knee and ankle joints were also in extension. There was no deformity observed overall as such. In general examination, he was afebrile, his pulse was 99 bpm, the respiratory rate was 36 bpm, blood pressure was 199/92 mmHg. There was evidence of Pallor and the capillary refilling time was more than 4 seconds. The inspection and palpatory findings divulged that patient is using accessory muscles for respiration. His precordium was found to be slightly flattened. The chest expansion was reduced at axillary and nipple level. The apex Impulse was heard at the fifth Intercostal Space. Pulsations were felt in the epigastric region. Parasternal Heave (Grade II according to AIIMS Grading) was palpated. A resonating note along with presence of thrill at 3<sup>rd</sup> and 4<sup>th</sup> intercostal space (Erb's area) during systole was found on percussion. On Auscultation, S1 & S2 (Soft S2) was heard. Single second heart sound heard: P<sub>2</sub> synchronous with A<sub>2</sub> (P<sub>2</sub>- closure of pulmonary valves. A<sub>2</sub>- Closure of Aortic valves) was heard & the murmur was pansystolic murmur. In Musculoskeletal system, there was no evidence of muscle wasting or atrophy. In Integumentary system evaluation, his skin was found to be pale and dry. There were no color changes in the cuticle, hyponychium or nail bed. There was no evidence of wounds or any swelling.

### Timeline:

19/01/2022	The patient was admitted.
21/01/2022	Diagnosed, planned for surgery, and was given Pre-operative Physiotherapy call.
25/01/2022	Operated with ICR-DCRV
28/01/2022	Shifted to the ward and continued Post-operative Physiotherapy.

**Table 1: Timeline**

**Diagnostic Assessment:****Diagnostic methods:**

**Blood Investigations:** The altered values than normal are given in Table 2.

	Results	Normal Values
<b>CBC: (Complete blood count)</b>		
Hemoglobin	9.4 mg/dL	11.2-14.5 mg/dL
Lymphocytes	55%	15-40%
<b>KFT: (Kidney function test)</b>		
Creatinine	0.3 mg/dL	0.8-1.2 mg/dL
<b>LFT: (Liver function test)</b>		
ALT	18 U/L	40 U/L
AST	44 U/L	40 U/L
<b>INR (International Normalized Ratio)</b>	0.9	1-1.3
<b>CKMB</b>	95 U/L	0-16 U/L

Table1: Altered values found in blood investigations.

**2D ECHO:**

Situs: Solitus.

Concordance: AV-VA concordance.

Relation of Great arteries: No relation of Great arteries.

Venous Drainage: 3PV-LA/ SP/ IVS RA.

Chambers: RA enlarged/ RVH – Mid. RV- Two turbulent jet shunts gradient of 48 mmHg.

Pulmonary Arteries: No supra-valvular membrane seen in PA.

IVS: No shunt seen over IVS.

Aorta and Arch: Right-sided Arch.No other shunt or lesion was seen.

Valves: Mitral, Aortic, Pulmonary & Tricuspid – Normal.

Final Diagnosis: Double Chambered right ventricle with a gradient of 48 mm of Hg.

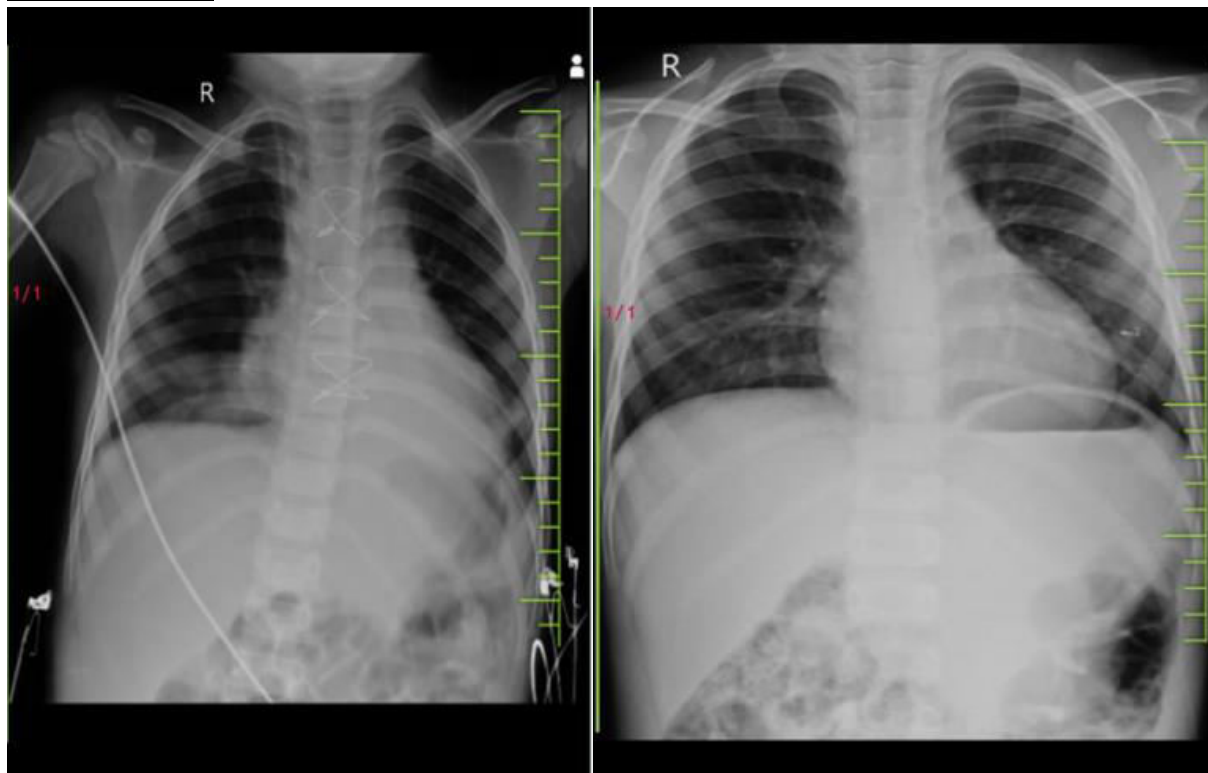
**Chest X-ray:**

Figure 1: Chest X-ray

**Diagnostic Challenges:**

Disease rarity, lack of biomarkers, asymptomatic patient till a latent phase, and lack of recent investigatory equipment at the patient's place were some of the diagnostic challenges in this case.

**Diagnosis:**

The patient was diagnosed with Double chambered right ventricle with Ventricular Septal defect by ruling out other conditions like Double outlet right ventricle, Double Inlet Right Ventricle, Tetralogy of Fallot, and Ebstein anomaly.


**Therapeutic Interventions:**

Owing to the individual variability of the disease onset, disease course, and disease progression, patients with DCRV will present with unique and different sets of symptoms thus, the interventions will vary. These children need structured care, not sympathy and that's exactly what a licensed physiotherapist can provide.

### **Patient-oriented Goals:**

- Patient and Parent Education about the importance of physiotherapy in the preoperative phase and treatment of effects of general anesthesia on lung function.
- Providing cost-effective treatment to cut off economic restraint.
- Prevent post-operative complications.
- Reduce unscheduled healthcare visits.
- Increase the potential level for daily physical activity.
- Reduce post-operative pain over the suture site from 6/10 to 3/10 in 2 weeks.
- Removal of lung secretions.
- Addressal of Breathing mechanics.
- Improve Exercise Tolerance.
- Preserve Pulmonary vasculature.
- Reduce Hospital Stay.
- Improve Core muscles Strength.
- Prevention of Infective Endocarditis.
- Improve the quality of life of the child.
- Ensure proper nutrition of the patient.
- Help the patient gain freedom and independence and make him liable to participate in recreational activities.

### **Physiotherapy Interventions:**

 Pre-operative:

- Breathing Exercises: **To preserve pulmonary vasculature.**
  1. Pursed Lip breathing: 20 repetitions x 1 set 5 times a day.
  2. Glossopharyngeal breathing: 20 repetitions x 1 set 5 times a day.
- Stretching exercises to:

Gastrocnemius, Soleus, Hamstrings, Tibialis Anterior, Quadriceps, Adductors, Quadratus Lumborum, Trapezius, Levator Scapulae, Flexor carpi ulnaris and radialis, Extensor carpi ulnaris and radialis, Biceps Brachii, Triceps Brachii, Trapezius, Levator Scapulae, Scalene muscles with 3 repetitions with 30 seconds hold in 1 set.

- Upper and lower Limb Mobility: To improve muscle strength.
  - In supine lying position:
    1. Ankle-Toe movements with hip and knee in extension: 20 repetitions x 1 set 3 times a day
    2. Hip-knee flexion ensuring that heel is being dragged over the bed instead of lifting: 10 repetitions x 1 set 3 times a day
    3. Static Quadriceps by placing pillow roll under heel: 10 repetitions x 1 set 3 times a day.
    4. Static Hamstrings by placing pillow roll under popliteal fossa: 10 repetitions x 1 set 3 times a day.
    5. Static Abdominals by placing pillow roll under the lordotic curvature of the lumbar spine: 10 repetitions x 1 set 3 times a day.
    6. Strengthening of Vastus Medialis Obliques by flexing both knees to 65 degrees and placing pillow roll between both knees: 10 repetitions x 1 set 3 times a day.
  - In Sitting over the edge of bed position:
    1. Dynamic Quadriceps by placing knees slightly forward to edge of bed 10 repetitions x 1 set 3 times a day.
    2. Range of motion exercises of wrist joint by placing a hand over pillow placed on patient's thighs.
    3. Ball squeezing activities.
    4. Elbow flexion-extension exercises.
  - In standing position:
    1. Shoulder flexion-extension, abduction-adduction, and internal-external rotation.
    2. Isometrics of the cervical spine.
    3. Range of motion exercises of Cervical Spine.
- Incentive Spirometry





#### Post-operative:

Treatment was done when the child was hemodynamically stable and never followed any potentially destabilizing maneuvers. Continuous observation of heart rate, blood pressure, pulmonary artery pressure, and oxygen saturation guided the progression of treatment.

- Breathing exercises.
- Strategies for Coughing with Splintage over suture site.
- Upper and lower Limb Mobility
- Incentive Spirometry
- Cryotherapy:
  - ✓ Started after 2weeks of surgery.
  - ✓ Application of Ice-packs by wrapping them in cotton napkin over suture site for 10-12 minutes for 4-5 hourly.
- Strategies to optimize trunk development included:
  - ✓ Changing the patient's body position frequently.
  - ✓ Improving the strength of weak pectoral muscles.
- Prevention of Valsalva Maneuver:
  - To prevent an increase in intrathoracic pressure.
  - ↓
  - To prevent a decrease in pulmonary flow.
  - ↓
  - Ensure proper circulation to the heart.
  - ↓
  - Maintain Cardiac output.
- ActiveCycle Breathing Technique:
 

The flexible method was adapted for mobilizing and clearing excess bronchial secretions. It consists of:

  1. Breathing Control:
 

The child was taken to a sitting position on a chair and asked to attain a calm state by relaxing his shoulders and performing diaphragmatic breathing at a normal pace (i.e., in his normal tidal volume) 4-5 times.
  2. Thoracic expansion exercises: **To decrease collapse of lung tissue and allow airflow via collateral channels- channels of Lambert and Martin & the pores of Kohn. The air behind secretions helps in mobilizing them through this**

**maneuver. Aids in lung expansion through the phenomenon of alveolar interdependence.**

- ✓ Deep Inspiration was followed by a 3-second hold prior to relaxed expiration.
- ✓ Another additional technique was adapted for proprioceptive stimulation by placing the physiotherapist's hand over the areas of the chest wall where the movement of the chest needs to be improved. This caused an initial improvement in ventilation to those areas and an additional increase in volume was achieved by teaching the child sniff maneuver at the end of deep inspiration.
- ✓ This was also combined with chest percussions, shaking, and vibrations to assist in the clearance of secretions.

**3. Forced Expiratory Exercises: Movement of peripherally situated secretions to central airways.**

This consisted of a combining one or two forced expirations (huffs) along with a phase of breathing control.

Huffing was taught at low volumes at the initial stage for more distally placed secretions to move more proximally. paroxysmal, huff at mid-lung volume was taught by instructing him to take a medium-sized breath with mouth and glottis open.

Precautions were taken to prevent continuous huffing for longer periods as it would have caused unnecessary paroxysmal coughing. It was also ensured that there were adequate pauses of 6-7 seconds for breathing control after 1-2 huffs to prevent any increase in airflow obstruction.

- Oral Suctioning.
- Prevention of Infective Endocarditis (IE):
  - ✓ Children with CHD have a greater risk of developing IE through infection caused by bacteria that enter the bloodstream and settle in the heart lining or a blood vessel, so the child must follow good dental hygiene that goes a long way towards preventing heart infection by reducing the risk of a tooth or gum infection.
  - ✓ The child and his parents were educated about the steps of good dental hygiene and its importance in preventing further complications. A few tips were:
    1. Brushing teeth with fluoride toothpaste twice a day. The child was advised to spit out the toothpaste and not to swallow it.
    2. Prevention of sipping sugary liquids to prevent tooth decay.

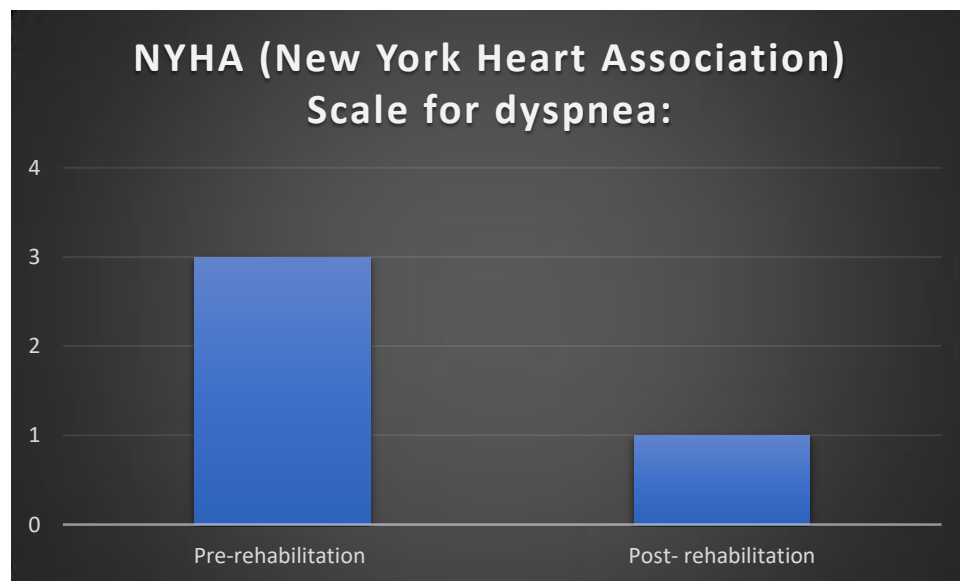
- Nutrition: To ensure proper muscle growth and development further preventing malnutrition.

A heart-healthy diet was prescribed to the child and advised parents to be taken care of:

- ✓ Eating vegetables and fruits as a whole daily.
- ✓ Use vegetable oil low in saturated fat and avoid butter or animal fats.
- ✓ Eating whole-grain, nuts, and seeds instead of bread, pastries, and fried chips.
- ✓ Fruits are full of Anthocyanins and flavonoids which are anti-inflammatory and regulate blood pressure like strawberries.
- ✓ Reducing intake of sweetened beverages and foods.
- ✓ Consumption of low-fat milk.
- ✓ Prevention of salty food items.

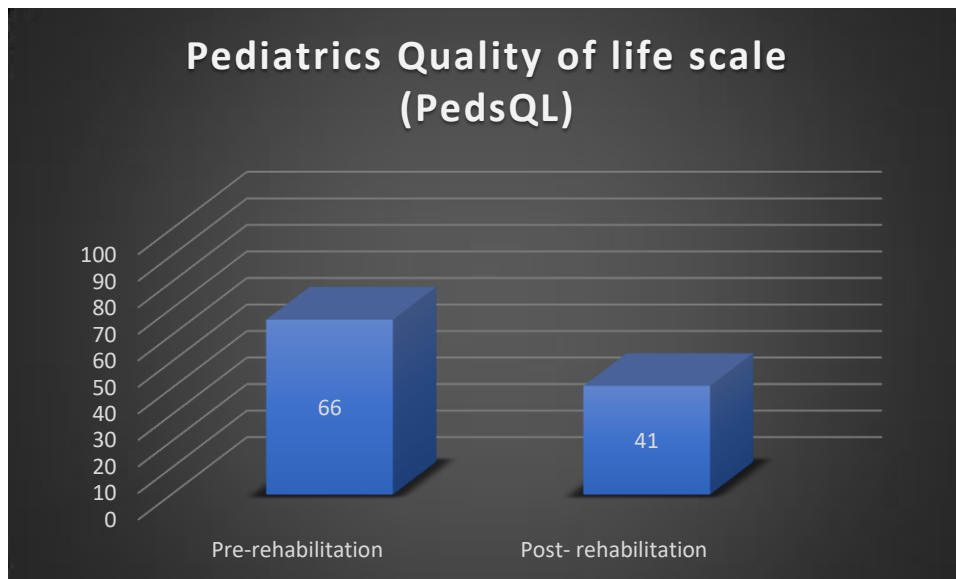
### Outcomes of intervention:

#### 1. NYHA (New York Heart Association) Scale for dyspnea:



Graph 1: NYHA

## 2. Pediatrics Quality of life scale (PedsQL):



**Graph 2: PedsQL**

### Discussion:

According to **Wong et al., (1991)**, DCRV is an acquired heart abnormality. It has not yet identified if there is any gene related propensity causing the aberrant muscle band development leading to such an aberration. Several DCRV mechanisms are proposed till date. Dislodgement of the septal marginal trabecula (moderator band) towards superior aspect has been hypothesized, specifically in the presence of a VSD and a turbulent flow in the RVOT. The turbulence flow was discussed by **Oliver et al., (2003)** which can cause DCRV by causing aberrant hypertrophy of the moderator band. This might help to explain the link between DCRV and VSD. **Darwazah et al., (2011)** provided information regarding the surgical intervention that is recommended in symptomatic or asymptomatic individuals when the peak gradient reaches 40 mm Hg. A right atriotomy, a right ventriculotomy, or a combination transarterial-transpulmonary access can all be used for excision of the aberrant muscle bands. The atriotomy of right side along with combination of transarterial-transpulmonary incision are the widely employed techniques. Because of the risk of ventricular arrhythmias, right ventriculotomy is only adapted in the most severe cases of RV dysfunction. Nonetheless, right ventriculotomy is still utilized in a small number of individuals with large and conspicuous bundles. The pentad of cases described demonstrates the wide range of DCRV manifestations, from a 'forme fruit' to extremely symptomatic. There was an accompanying PM-VSD in all patients, as well as aberrant muscle bundles in the RVOT. Shortness of breath and chest discomfort was the primary presenting symptoms in the first patient. She was originally asymptomatic due to a tiny isolated VSD

diagnosed early in infancy. However, in a very short amount of time, she experienced increasing symptoms with a considerable gradient across the RVOT, necessitating surgical correction. Later in life, the second sufferer had shortness of breath. She started to be asymptomatic with a solitary VSD identified at the age of 30. Despite this, she experienced worsening symptoms with a large gradient across the RVOT, necessitating surgical correction. The third patient was found to be born with patent ductus arteriosus, which was corrected early in life with surgical ligation, and a minor VSD. She was asymptomatic when she was diagnosed with DCRV, and there was no major gradient over the RVOT. The fourth patient being born with VSD, required surgical intervention at the age of 11 years. He was not having any symptoms until he acquired DCRV following closure for VSD, and there was no substantial gradient over RVOT. The fifth patient presented with no symptoms and was initially diagnosed with a single VSD. He was then diagnosed with DCRV without discernible gradient over the RVOT. The case series supports this idea that relative aggravating blockage in DCRV, along with VSD, is a procured condition with hereditary predisposition.

**Hoffman et al., (2004)** discovered that the DCRV's incidence varies from 0.5% to 2%. **Alva et al., (1999)** stated that it is connected with VSD in 80% to 90% of instances of Subaortic stenosis, pulmonary valve stenosis, double-outlet RV, tetralogy of Fallot, abnormal pulmonary venous drainage, complete or corrected transposition of the great veins, pulmonary atresia with an intact septum, and Ebstein anomaly are some of the other concomitant disorders. **Cil et al., (1995)** told that DCRV is more common among children, according to research. The average age of presentation ranges from 4 months to 20 years. The vast majority of individuals are asymptomatic and are referred for heart murmur examination. In a study of 52 individuals, Cil and colleagues discovered that 40% were asymptomatic, 35% were fatigued, and 17% had exertional dyspnea; congestive heart failure, cyanosis, and palpitation were present in 10% to 12% of the same sample. **Pongiglione et al., (1982)** showed that the patients they represent had quite diverse DCRV symptoms. They differ in terms of presentation, location of aberrant obstructive muscle bundles, and the existence of accompanying lesions. Although inpatient 1 showed various indications of congestive heart failure, his major presenting symptom was chest discomfort. The aberrant muscle bundle was fibromuscular, distinct, high, and horizontal in appearance. The absence of a cardiac lesion was established intraoperatively. The first murmur was identified in childhood and was considered to be a VSD, however, it was always an isolated DCRV. This explains our patient's excruciating symptoms. Patient 2 was asymptomatic, which is unusual in this age range. The low-pressure gradient and tiny VSD account for the lack of symptoms. This example corroborated Pongiglione and colleagues' idea that blockage by abnormal muscular bundles is an acquired characteristic in VSD patients. **Nagashima et al., (2005)** indicated that all symptomatic adult patients should be operated on to resect the obstructive muscle bundles and correct any related lesions. Because the blockage in adults progresses fast, this also applies to asymptomatic individuals with gradients greater than 40 mmHg. The anomalous muscle is possible to be resected by a right atriotomy, a right ventriculotomy, or a combination transarterial–transpulmonary incision.

### **Conclusion:**

Cardiovascular and pulmonary physiotherapy is an evidence-based practice that evolved rapidly in a few decades and works parallel to the medical and surgical team's intervention. It is a cost-effective form of treatment that rapidly helps in the betterment of patients suffering from pulmonary as well as cardiac illnesses. A Cardiovascular & Pulmonary physiotherapist's training provides a good foundation for integrative engagement in multidisciplinary cardiac rehabilitation.

### **Acknowledgment:**

We hereby thank all the authors and guides for their contribution in the form of immense support, tutoring, and briefing for the preparedness of this case report.

### **Patient's Perspective:**

As the parents reported, "Our child was able to cope with the physical demands imposed on him during playing and strenuous activities compared to before. He is more cheerful and able to manage his daily chores by himself. He stays motivated most of the time for the physiotherapy sessions as he understands its vital role in the betterment of his condition."

### **Competing Interests:**

The authors declare no competing interests as such.

### **Authors' contributions:**

The design of this report was suggested by TB, PB & AS. The study was made and implemented with the help of TB, PB, VY & AS. TB wrote this report. The final report was analyzed and approved for publishing by VY.

### **Informed Consent:**

The patient & parents were informed about the study followed by obtaining oral informed consent.

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