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RESEARCH ARTICLE

CHYLOTHORAX FOLLOWING BIDIRECTIONAL CAVOPULMONARY SHUNT (GLENN PROCEDURE) IN AN INFANT WITH COMPLEX CONGENITAL HEART DISEASE: A CASE REPORT

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Abstract

Background: Chylothorax is a serious complication that may occur after pediatric cardiac surgery. We report the case of a 17-month-old infant with complex congenital heart disease of the single-ventricle type, who developed recurrent massive pleural effusion after a bidirecti onal cavopulmonary shunt (Glenn procedure). This case illustrates a recognized complication that may be difficult to manage.

Introduction:-

The bidirectional cavopulmonary shunt (Glenn procedure) represents an intermediate surgical step in the staged palliation of single-ventricle congenital heart disease. It enables passive pulmonary circulation via the superior vena cava [1]. However, this non-pulsatile circulation predisposes patients to specific complications, including pleural effusions, chylothorax, and persistent hypoxemia. These complications are reported in approximately 30% of postoperative cases [2,3]. We report a case of chylothorax causing acute respiratory distress in an infant following a bidirectional cavopulmonary shunt.

Case Report:-

Patient demographics and medical history:-

We report the case of a 17-month-old male infant, the youngest of four siblings, born at term following a poorly monitored pregnancy, with no parental consanguinity. He had been followed since the age of one year for complex congenital heart disease comprising: single ventricle, mitral valve atresia, right-sided aorta, sub-valvular and valvular pulmonary stenosis, and an interatrial communication of 7 mm. One month prior to admission, the patient underwent a bidirectional cavopulmonary shunt (Glenn procedure). The immediate postoperative course was uneventful, with good hemodynamic stability, and pulmonary arterial pressure increased from 10 mmHg to 15 mmHg.

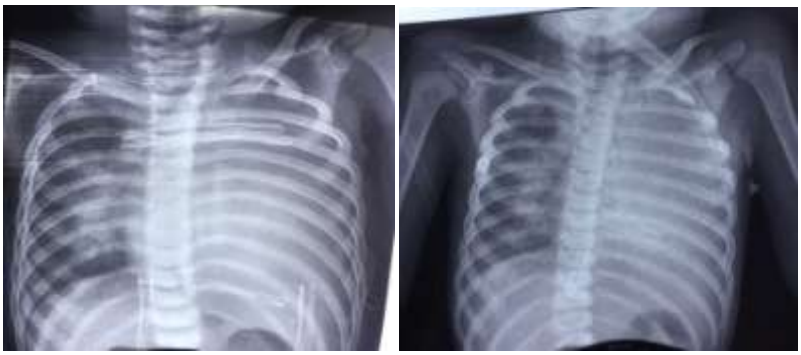
History of presenting illness:-

The illness began 15 days before admission with the gradual onset of afebrile respiratory distress. Chest imaging revealed a large left-sided pleural effusion, which prompted a first thoracocentesis. The course was complicated by recurrence one week later, requiring a second drainage procedure that yielded approximately 600 mL of purulent-

appearing fluid. Given the persistence of symptoms, the patient was referred to the Pediatric Intensive Care Unit (PICU) for further management.

Physical examination on PICU admission:-
On admission, clinical examination revealed preserved consciousness (GCS 15/15), tachycardia at 154 bpm, tachypnea at 40 cycles/min, SpO_2 at 65% on supplemental oxygen, peripheral cyanosis, and signs of increased work of breathing (intercostal retractions, nasal flaring).

Investigations:-
Laboratory investigations are summarized in Table 1.
Chest X-ray showed persistent left-sided pleural effusion.



Pleural fluid analysis (Table 2) confirmed the diagnosis:

Table 1: Laboratory Investigations on PICU Admission

Parameter	Result
Hemoglobin	11.7–13.7 g/dL
White blood cells	11,980/mm³
Platelets	405,000/mm³
CRP	6.3 mg/L
Na^+	134 mmol/L
K^+	3.1–3.8 mmol/L

Table 2: Pleural Fluid Biochemical Analysis

Parameter	Result
Total proteins	46 g/L (exudative)
Triglycerides	2.68 mmol/L (≈ 237 mg/dL) — diagnostic threshold: ≥ 1.24 mmol/L
Cytobacteriological examination	Sterile

A triglyceride level of 2.68 mmol/L (237 mg/dL), well above the diagnostic threshold of 1.24 mmol/L (110 mg/dL), confirmed the diagnosis of chylothorax. The high protein content was consistent with an exudative effusion. Sterile cytobacteriological examination excluded secondary infection.

Management and outcome:-
In the PICU, the patient received furosemide 1 mg/kg every 6 hours to reduce systemic venous pressure and lymphatic flow, aspirin 20 mg/kg/day for its antiplatelet and anti-inflammatory properties in the context of cavopulmonary circulation, and continuous chest tube drainage. The clinical course was favorable, with progressive respiratory improvement allowing transfer to a non-intensive care ward.

Discussion:-

Epidemiology:-
Chylothorax is one of the most feared postoperative complications in pediatric cardiac surgery. According to North American multicenter databases (PC4 and PHIS), the overall incidence ranges from 2.8% to 3.8% [1]. This incidence is significantly higher in neonates (6.9%), patients with single-ventricle physiology (6.9%), chromosomal anomalies (5.2%), and major non-cardiac anomalies (6.4%) [4]. A recent study using the US National Inpatient Sample (2016-2019) confirmed that cavopulmonary shunting (Glenn and Fontan) carries the highest postoperative chylothorax incidence among all cardiac surgeries [5]. In single-ventricle series, chylothorax complicates up to 17% of Glenn procedures, particularly in patients with pre-existing lymphatic abnormalities on MRI [3]. The presence of a systemic right ventricle has been identified as an independent risk factor, owing to ventricular dysfunction and elevated central venous pressure [6].

Diagnosis:-
Diagnosis is based on pleural fluid biochemistry. A triglyceride level >110 mg/dL (1.24 mmol/L) with cholesterol <200 mg/dL (5.18 mmol/L) is considered diagnostic, corresponding to a 99% diagnostic probability [7]. In our case, triglycerides measured 2.68 mmol/L (237 mg/dL), well above threshold. The initially 'purulent' appearance of the fluid underscores the importance of systematic biochemical analysis in any recurrent post-Glenn effusion.

Management:-
Initial management follows a stepwise conservative approach, guided by the 2022 PC4 Chylothorax Work Group multicenter consensus algorithm [8]. Dietary modification with a medium-chain triglyceride (MCT) diet is the cornerstone; a two-week duration has been shown to reduce drainage duration without increasing recurrence risk [9]. Furosemide reduces systemic venous pressure and lymphatic flow. Chest tube drainage is indicated for large effusions with respiratory compromise. Octreotide should be introduced in case of treatment failure or recurrence [8,10]. In our case, the combination of chest drainage, furosemide, and aspirin resulted in a favorable outcome without requiring somatostatin analogues.

Conclusion:-

This case illustrates the pathophysiological and therapeutic complexity of post-Glenn chylothorax. Knowledge of its epidemiology (6.9% incidence in single-ventricle patients), its mechanisms (thoracic duct injury, venous hypertension, pre-existing lymphatic dysfunction), and the updated management algorithm (MCT diet, diuretics, octreotide in case of failure [8,10]) is essential for any clinician managing complex congenital heart disease. Systematic biochemical analysis of all pleural effusions following cavopulmonary surgery, application of a standardized stepwise protocol and early identification of risk factors are key determinants of a favorable outcome.

Declarations:-

Patient Consent: Written informed parental consent was obtained for publication of this case report and the accompanying images.

Conflict of Interest: The authors declare that there is no conflict of interest.

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Authors' Contributions: All authors contributed to the clinical management of the patient and to the drafting and critical revision of the manuscript. All authors approved the final version.

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