



Evaluation of pulmonary arteries and source of pulmonary blood supply in children with pulmonary atresia and ventricular septal defect using CT angiography

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ABSTRACT

Objective: To evaluate pulmonary arteries and determine the sources of pulmonary blood supply in children with pulmonary atresia and ventricular septal defect (PA-VSD) using computed tomography angiography (CTA).

Methods: This retrospective cross-sectional study was conducted at Pediatric Cardiology Clinic, Hayatabad Medical Complex, Peshawar, Pakistan, from January 2009 to December 2019. Pediatric patients (≤ 18 years) with confirmed PA-VSD who underwent CTA for preoperative anatomical assessment were included. CTA was used to evaluate pulmonary artery morphology, patent ductus arteriosus (PDA), and major aortopulmonary collateral arteries (MAPCAs). Based on CTA findings, patients were classified according to the source of pulmonary blood supply into Type A (native pulmonary arteries supplied via PDA), Type B (native pulmonary arteries supplied by both PDA and MAPCAs), and Type C (absence of native pulmonary arteries with MAPCAs only). Statistical analysis was performed using SPSS version-22.

Results: Ninety-one patients were included, with a mean age of 3.2 ± 2.8 years; 58 (63.7%) were males. Cyanosis was the most common presenting symptom (94.5%). Type B PA-VSD was the most frequent anatomical pattern (46.2%), followed by Type C (27.5%) and Type A (26.4%). Twenty-four patients (26.4%) had oxygen saturations below 60%, 50 patients (54.9%) had oxygen saturations between 60% and 80%, and 17 patients (18.7%) had oxygen saturations above 80% ($p=0.018$). CTA demonstrated higher detection rates than echocardiography for MAPCAs (83.3% vs 47.4%, $p<0.001$) and pulmonary artery stenosis (25.6% vs 10.3%, $p=0.012$).

Conclusion: CTA provides comprehensive anatomical delineation of pulmonary arteries and collateral circulation in PA-VSD and is valuable for accurate preoperative evaluation.

Keywords: Computed Tomography Angiography (MeSH); Diagnostic Imaging (MeSH); Pulmonary Atresia (MeSH); Heart Septal Defects, Ventricular (MeSH); Child (MeSH); Heart Defects, Congenital (MeSH); Pulmonary Circulation (MeSH); Heart Septal Defects (MeSH); Aortopulmonary Septal Defect (MeSH); Major Aortopulmonary Collateral Arteries (Non-MeSH); Pulmonary Artery (MeSH).

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INTRODUCTION

Pulmonary atresia with ventricular septal defect (PA-VSD) represents a complex and heterogeneous form of cyanotic congenital heart disease characterized by the absence of luminal continuity between the right ventricle and the pulmonary artery. In this condition,

pulmonary blood flow is not directly supplied from the right ventricle but instead occurs through a ventricular septal defect, allowing systemic-to-pulmonary circulation.¹ PA-VSD encompasses a broad spectrum of anatomical variations, and its management requires individualized treatment strategies based on detailed characterization of the pulmonary

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vasculature, the source of pulmonary blood supply, and the presence or absence of major aortopulmonary collateral arteries (MAPCAs).^{2,3}

From a developmental and morphological perspective, PA-VSD is often considered an extreme variant of classic Tetralogy of Fallot, in which right ventricular outflow tract obstruction manifests as complete atresia rather than stenosis. The condition retains the other classical components of Tetralogy of Fallot, including a maligned ventricular septal defect, an overriding aorta, and right ventricular hypertrophy. Congenital heart diseases (CHDs) are among the most common congenital anomalies, affecting approximately 1% of live births, corresponding to about 6.7 cases per 1,000 live births.⁴ Within this group, PA-VSD is relatively uncommon, accounting for approximately 0.2% of live births and nearly 2% of all congenital heart defects.⁵ A key feature of PA-VSD is the marked variability in pulmonary blood supply, which has important implications for clinical management and surgical planning. Accurate anatomical characterization of the pulmonary arterial system and collateral circulation is therefore essential.⁶ A contemporary classification system categorizes PA-VSD (Figure 1) into three principal types according to the pattern of pulmonary blood supply. Type A includes cases in which pulmonary circulation is supplied exclusively through native pulmonary arteries (NPA). Type B represents a dual-supply pattern in which both NPA and MAPCAs contribute to pulmonary

perfusion. Type C is characterized by the complete absence of native pulmonary arteries, with pulmonary blood flow provided entirely through MAPCAs.⁷ This classification framework facilitates standardized anatomical reporting and supports surgical decision-making. In addition to defining pulmonary circulation patterns, comprehensive preoperative evaluation must also identify associated cardiovascular anomalies, such as partial anomalous pulmonary venous connections and coronary-to-pulmonary artery fistulae. These associated abnormalities may significantly influence surgical strategy and overall clinical outcomes.⁸ Transthoracic echocardiography is generally the first-line imaging modality for the evaluation of congenital heart disease and provides reliable information regarding intracardiac anatomy. However, its ability to delineate extracardiac vascular structures, particularly complex pulmonary arterial anatomy and collateral vessels, may be limited by suboptimal acoustic windows. Cardiac catheterization has traditionally been regarded as the reference standard for detailed vascular assessment; nevertheless, it is invasive, associated with radiation exposure and contrast administration, and carries inherent procedural risks. Computed tomography angiography (CTA) has

recently emerged as a valuable non-invasive imaging modality for the comprehensive assessment of cardiovascular anatomy in patients with PA-VSD. CTA provides high-resolution visualization of both intracardiac and extracardiac structures, enabling accurate delineation of pulmonary arteries, MAPCAs, and associated vascular anomalies, while avoiding the invasiveness of catheter-based procedures.^{9,10}

In the evolving landscape of pediatric cardiology in our region, the incorporation of CTA into the diagnostic work-up of complex congenital heart disease represents a relatively recent development. The present study was planned to evaluate the role of CTA in defining the detailed cardiac and vascular anatomy of pediatric patients with pulmonary atresia and ventricular septal defect, thereby facilitating improved surgical planning and optimizing clinical outcomes.

METHODS

This retrospective cross-sectional study was conducted at the Pediatric Cardiology Clinic of Hayatabad Medical Complex, Peshawar, Pakistan, from January 2009 to December 2019. Ethical approval for the study was obtained from the Institutional Review

Board of Hayatabad Medical Complex (Reference #: 1720; dated: January 11, 2024). Written informed consent was obtained from the parents or legal guardians of all participants prior to CT angiography.

The inclusion criteria comprised pediatric patients (≤ 18 years) with a confirmed diagnosis of PA-VSD who underwent CT angiography for preoperative anatomical evaluation and had imaging studies of adequate diagnostic quality. Patients were excluded if CT angiography studies were incomplete, if image quality was compromised by significant motion artefacts or inadequate contrast opacification that hindered anatomical assessment, or if clinical records were incomplete. All eligible patients presenting with PA-VSD during the study period were consecutively enrolled. A total of 120 patients were initially identified, of whom 91 met the eligibility criteria and were included in the final analysis.

Patient demographic and clinical data, including age at imaging, gender, body weight, body surface area, and baseline oxygen saturation in room air (SpO_2), were retrieved from medical records. All CT angiographic examinations were performed using a 64-slice multidetector CT scanner (Aquilion 64, Toshiba Medical Systems, Japan). Non-

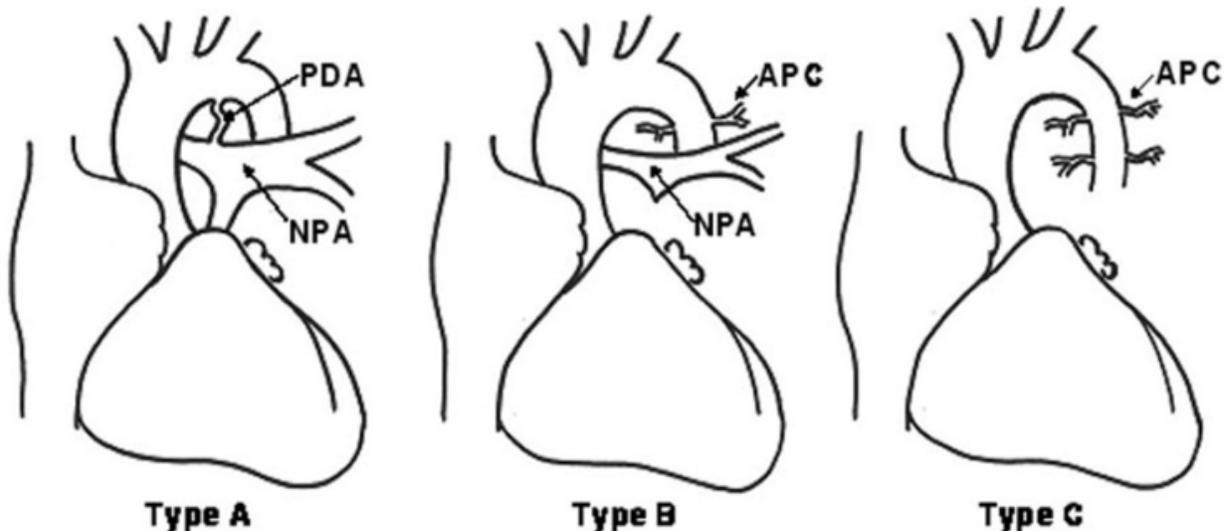


Figure 1: Classification of PA - VSD according to the status of native pulmonary arteries, Major aorto-pulmonary collaterals (APCs). Type A: Native pulmonary arteries present, no APCs, Type B: Native pulmonary arteries and APCs present, Type C: No native pulmonary arteries, only APCs maintain pulmonary blood flow.⁷

ionic contrast medium with a concentration of 300 mg/ml was administered intravenously at a dose of 1.5-2 ml/kg. Image reconstruction was performed with axial slices at a thickness of 0.625 mm. Multiplanar reformatted (MPR) images were generated in the coronal and sagittal planes, along with maximum intensity projection (MIP) images and three-dimensional volume-rendered (3D-VR) reconstructions to facilitate detailed visualization of cardiovascular structures.

All CT angiographic studies were independently reviewed and interpreted by a board-certified pediatric cardiologist with more than 10 years of experience in cardiovascular imaging. Image analysis was performed on a dedicated workstation using Toshiba Aquilion 64 software (version 3.0). The anatomical evaluation included assessment of native pulmonary arteries, their morphology and dimensions; identification and characterization of the patent ductus arteriosus (PDA); and evaluation of major aortopulmonary collateral arteries (MAPCAs). Additional parameters assessed included the location and size of the ventricular septal defect, aortic arch sidedness and branching pattern, and the presence of associated cardiovascular anomalies.

Based on CT angiographic findings, patients were classified according to the source of pulmonary blood supply into three groups: Type A, presence of native pulmonary arteries supplied by a PDA without MAPCAs; Type B, native pulmonary arteries with dual blood supply from both the PDA and MAPCAs; and Type C, absence of native pulmonary arteries with pulmonary perfusion supplied exclusively by MAPCAs.

Statistical analysis was performed using SPSS version 22 and Microsoft Excel version 19. Continuous variables were expressed as mean ± standard deviation (SD), whereas categorical variables were presented as frequencies and percentages. Differences among PA-VSD types were analyzed using the chi-square test for categorical variables and one-way analysis of variance (ANOVA) for continuous variables. McNemar's

test was used to compare findings between echocardiography and CT angiography. A p-value of <0.05 was considered statistically significant.

RESULTS

A total of 91 patients with PA-VSD who underwent CT angiography were included in the final analysis. The mean age at imaging was 3.2±2.8 years (range: 2 months to 16 years), with 58 patients (63.7%) males and 33 (36.3%) females. The mean body weight was 12.4±7.6 kg, and the mean body surface area was 0.54±0.28 m². The overall mean oxygen saturation in room air was 69.8±12.4% (range: 45-92%). Clinical presentation varied, with cyanosis the most common presenting symptom in 86 patients (94.5%), followed by dyspnea on exertion in 62 patients (68.1%). The demographic and clinical characteristics are summarized in Table I.

Table II summarizes the distribution of clinical and anatomical variables across PA-VSD types. No significant association was found between gender

and PA-VSD type (p=0.542). Similarly, there was no significant difference in mean age at presentation among the three types (p = 0.416). Based on CTA findings, Type B pulmonary atresia (both native pulmonary arteries and MAPCAs present) was the most common, identified in 42 patients (46.2%), followed by Type C (MAPCAs only) in 25 patients (27.5%), and Type A (native pulmonary arteries only) in 24 patients (26.4%). A statistically significant association was observed between oxygen saturation levels and PA-VSD type (p = 0.004). Patients with Type A pulmonary atresia had significantly higher mean oxygen saturation (76.2±9.8%) compared to Type B (68.4±11.6%) and Type C (65.8±13.2%). No significant association was found between gender and PA-VSD type (p=0.542). Similarly, there was no significant difference in mean age at presentation among the three types (p=0.416). Table III compares echocardiographic and CTA findings in the evaluation of key anatomical features in PA-VSD. CTA demonstrated significantly higher detection rates for

Table I: Demographic and clinical characteristics of patients (n=91)

| Variables | | Values |
|-------------------------------------|---------------------|-------------|
| Age (years) | Mean ± SD | 3.2 ± 2.8 |
| | Range | 0.17-16 |
| Gender, n (%) | Male | 58 (63.7) |
| | Female | 33 (36.3) |
| Body Weight (kg) | Mean ± SD | 12.4 ± 7.6 |
| Body Surface Area (m ²) | Mean ± SD | 0.54 ± 0.28 |
| Oxygen Saturation (%) | Mean ± SD | 69.8 ± 12.4 |
| | <60%, n (%) | 24 (26.4) |
| | 60-80%, n (%) | 50 (54.9) |
| | >80%, n (%) | 17 (18.7) |
| Aortic Arch Position, n (%) | Left | 64 (70.3) |
| | Right | 27 (29.7) |
| Clinical Presentation, n (%) | Cyanosis | 86 (94.5) |
| | Dyspnea on exertion | 62 (68.1) |
| | Failure to thrive | 34 (37.4) |
| | Heart failure | 18 (19.8) |

Table II: Distribution of PA-VSD types according to clinical and anatomical variables

| Variables | | Type A (n=24) | Type B (n=42) | Type C (n=25) | p-value |
|------------------------------------|--------|---------------|---------------|---------------|---------|
| Gender, n (%) | Male | 14 (58.3) | 28 (66.7) | 16 (64.0) | 0.542† |
| | Female | 10 (41.7) | 14 (33.3) | 9 (36.0) | |
| Age (years), Mean±SD | | 2.8±2.4 | 3.6±3.1 | 2.9±2.5 | 0.416‡ |
| Oxygen Saturation (%), Mean±SD | | 76.2±9.8 | 68.4±11.6 | 65.8±13.2 | 0.004‡ |
| SpO ₂ Categories, n (%) | <60% | 2 (8.3) | 12 (28.6) | 10 (40.0) | 0.018† |
| | 60-80% | 13 (54.2) | 24 (57.1) | 13 (52.0) | |
| | >80% | 9 (37.5) | 6 (14.3) | 2 (8.0) | |
| Aortic Arch, n (%) | Left | 19 (79.2) | 28 (66.7) | 17 (68.0) | 0.328† |
| | Right | 5 (20.8) | 14 (33.3) | 8 (32.0) | |

†Chi-square test; ‡One-way ANOVA; †Statistically significant (p<0.05); PA-VSD: Pulmonary atresia and ventricular septal defect; SpO₂: Peripheral capillary oxygen saturation

Table III: Comparative analysis of echocardiographic and CTA findings

| Parameters | Echocardiography | CTA | Agreement (%) | p-value |
|-----------------------------------|------------------|--------------|---------------|---------------------|
| Native PA Identification, n/N (%) | 70/78 (89.7) | 78/78 (100) | 89.7 | 0.003 [†] |
| MAPCA Detection, n/N (%) | 37/78 (47.4) | 65/78 (83.3) | 56.4 | <0.001 [†] |
| PA Stenosis Detection, n/N (%) | 8/78 (10.3) | 20/78 (25.6) | 84.6 | 0.012 [†] |
| Aortic Arch Position, n/N (%) | 78/78 (100) | 78/78 (100) | 100 | 1.000 |

†Statistically significant (p < 0.05), PA, pulmonary artery; MAPCA, major aortopulmonary collateral artery; CTA, computed tomography angiography

native pulmonary arteries (100% vs 89.7%, p=0.003), MAPCAs (83.3% vs 47.4%, p<0.001), and pulmonary artery stenosis (25.6% vs 10.3%, p=0.012) compared with echocardiography. In contrast, both imaging modalities showed complete agreement in identifying the aortic arch position (100%, p=1.000).

DISCUSSION

This retrospective study highlights the diagnostic utility of CTA in the comprehensive anatomical evaluation of pulmonary arteries and pulmonary blood supply in pediatric patients with PA-VSD. The findings emphasize the importance of advanced cross-sectional imaging in preoperative assessment and surgical planning for this complex congenital cardiac anomaly. A male predominance (63.7%) was observed in our cohort, which is consistent with previous reports showing a male-to-female ratio ranging from 1.3:1 to 2:1.¹¹ Cyanosis was the predominant presenting feature (94.5%), reflecting the fundamental pathophysiology of PA-VSD in which systemic venous blood is

diverted into the systemic circulation through a ventricular septal defect due to the absence of right ventricular outflow to the pulmonary arteries. The mean oxygen saturation of 69.8% in our cohort is comparable to values reported in untreated PA-VSD populations.¹²

Regarding anatomical classification, Type B PA-VSD was the most common variant (46.2%), followed by Type C (27.5%) and Type A (26.4%), a distribution consistent with international registry data.^{6,13} A significant association was observed between PA-VSD type and oxygen saturation (p=0.004). Patients with Type A disease demonstrated higher mean oxygen saturation (76.2%) compared with Type B (68.4%) and Type C (65.8%), suggesting that native pulmonary arteries provide more stable pulmonary perfusion than collateral-dependent circulation.^{14,15} Furthermore, the proportion of patients with severe hypoxemia increased progressively from Type A (8.3%) to Type B (28.6%) and Type C (40.0%), which may have implications for the timing of surgical intervention in patients with advanced

cyanosis.¹⁶

The left aortic arch was the predominant anatomical configuration (70.3%), although the prevalence of right aortic arch (29.7%) was higher than that observed in the general population, supporting the known association between PA-VSD and aortic arch laterality anomalies.¹⁷ Comparative analysis between echocardiography and CTA revealed significant differences in the detection of extracardiac vascular structures. CTA identified MAPCAs more frequently than echocardiography (83.3% vs 47.4%, p<0.001) and also demonstrated higher detection rates for pulmonary artery stenosis (25.6% vs 10.3%, p=0.012). These findings are consistent with previous studies showing the superiority of cross-sectional imaging for evaluating complex vascular anatomy in congenital heart disease.¹⁸ The relatively low sensitivity of echocardiography for MAPCA detection (56.4%) reflects the limitations of ultrasound in visualizing tortuous collateral vessels arising from the descending thoracic aorta. A study

from Zurich involving 98 patients reported a similar echocardiographic sensitivity of approximately 53% for MAPCA identification.¹⁹ Additionally, pulmonary artery stenosis was underestimated by echocardiography, with approximately 15.4% of cases missed, which may have important implications for surgical planning.²⁰

The detailed anatomical delineation provided by CTA has direct implications for operative decision-making. The classification of PA-VSD into Types A, B, and C determines the overall surgical strategy: Type A patients are generally suitable for complete repair with restoration of right ventricle-to-pulmonary artery continuity; Type B patients often require rehabilitation of MAPCAs combined with augmentation of native pulmonary arteries; whereas Type C patients typically undergo staged unifocalization procedures.²¹ By providing high-resolution visualization of pulmonary arteries, MAPCAs, and associated anomalies, CTA facilitates precise surgical planning and allows anticipation of technical challenges during repair.²² In addition, the identification of associated abnormalities, such as coronary artery anomalies, is essential to avoid potential intraoperative complications.²³

Limitations and future directions

Several limitations warrant acknowledgment. The retrospective, single-center design may introduce selection bias and limits generalizability. Inter-observer variability in CTA interpretation was not formally assessed. The lack of surgical or catheterization correlation for all cases prevents comprehensive validation against alternative reference standards, and long-term outcome data were not available. Emerging technologies including three-dimensional flow MRI and prospective multicenter registries correlating detailed anatomical characterization with surgical outcomes would provide valuable insights into optimal treatment algorithms.

CONCLUSION

This study demonstrates that CTA provides comprehensive, reliable anatomical characterization of

pulmonary arteries and pulmonary blood supply in children with PA-VSD. CTA demonstrates superior diagnostic performance compared to echocardiography for detection of major aortopulmonary collateral arteries and pulmonary artery stenosis. Type B PA-VSD represents the most common anatomical configuration, and oxygen saturation correlates significantly with PA-VSD type. The integration of CTA into the diagnostic algorithm enables optimized preoperative assessment and contributes to improved surgical planning in this complex congenital cardiac malformation.

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AUTHORS' CONTRIBUTION

The following authors have made substantial contributions to the manuscript as under:

ZU: Conception and study design, critical review, approval of the final version to be published

KK & YR: Acquisition, analysis and interpretation of data, drafting the manuscript, approval of the final version to be published

Authors agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

CONFLICT OF INTEREST

Authors declared no conflict of interest, whether financial or otherwise, that could influence the integrity, objectivity, or validity of their research work.

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DATA SHARING STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.



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