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RESEARCH ARTICLE

Evaluation of Tracheobronchial Compressions due to Cardiovascular Diseases

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ABSTRACT

Objective: Cardiovascular system-associated tracheobronchial compressions are often unrecognized complications of congenital cardiac anomalies, which may complicate the management of the disease. Enlarged or malpositioned cardiovascular anatomy or vascular rings can cause airway compression.

Materials and Methods: The study included patients who were followed up with a diagnosis of cardiovascular system-associated tracheobronchial compression during a two-year period at a tertiary center. Clinical data and short-term outcomes were retrospectively evaluated.

Results: The study included 22 patients with a median age of 24 months (IQR 11–29 months); 68.2% (n=15) were male. The median age at diagnosis was 15 days (IQR 0–18 months). Recurrent wheezing (59.1%) and dyspnea (59.1%) were the most common symptoms. There were 10 patients (45.5%) in the vascular ring/pulmonary sling group, while 12 patients (54.5%) were in the abnormally enlarged or malpositioned cardiovascular structure due to the underlying congenital heart disease group. The trachea was the most frequently compressed airway structure, with a frequency of 68.2%, while the left main bronchus followed with a frequency of 40.9%. The median age at surgical intervention was 5.5 months (IQR 2.25–9 months), while only two patients were managed conservatively. The overall mortality rate was 4.5% during follow-up.

Conclusion: A high index of suspicion and early diagnosis are essential for the optimal management of patients with cardiovascular system-associated airway compressions. Long-term studies are needed in this area to standardize diagnostic and therapeutic management.

Keywords: Congenital heart disease, stridor, tracheobronchial compression, vascular ringwheezing

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INTRODUCTION

The tracheobronchial and cardiovascular systems are closely related to each other both physiologically and anatomically. An abnormal anatomical structure in the cardiovascular system can cause compression in the tracheobronchial system.[1] Even though survival of patients with congenital heart diseases has increased during recent years due to advances in surgical techniques and improved postoperative medical and respiratory care facilities, comorbidities, including tracheobronchial compressions, can make the management of the disease highly difficult. The prevalence of tracheobronchial compressions in children with congenital cardiovascular diseases undergoing surgery is reported to be 1–2%;[2] however, the exact prevalence may be higher due to underrecognition and diagnostic challenges.

The etiology of vascular compression of the tracheobronchial system can be evaluated in two main groups: vascular rings/pulmonary artery sling and malpositioned or

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enlarged cardiovascular anatomy due to underlying congenital heart disease.[2,3] Vascular rings are the most common causes of tracheobronchial compression, and they may cause extrinsic airway malacia.[4] Dilated pulmonary arteries due to complex congenital heart diseases with left-to-right shunt, tetralogy of Fallot, truncus arteriosus, and malpositioned or dilated aorta are some of the other reasons that may also cause airway compression.[5]

Symptoms and age at presentation depend on the severity and location of the vascular compression.[1] The trachea and main bronchus are the most frequently affected structures.[5] Some children with severe airway compression may require ventilation support for a long time. Milder forms of airway compression may be asymptomatic or present with a variety of nonspecific symptoms, including cough, wheezing, dyspnea, stridor, dysphagia, recurrent respiratory infections, and apneas, which makes diagnosis challenging. Asthma is a common misdiagnosis, especially for older children.[3] A high index of suspicion is required as the symptoms are often nonspecific.[6]

Diagnosis typically involves a combination of imaging modalities. Echocardiography is essential for the evaluation of congenital heart diseases and usually shows abnormal vascular structures clearly; however, evaluation of airway compression is limited. Chest radiography and barium swallow studies have been replaced by multidetector computerized tomography (MDCT) and magnetic resonance imaging. MDCT is the primarily preferred method in most centers, with improved multiplanar reconstruction technologies, increased speed, and modified protocols with reduced radiation dose. Flexible fiberoptic bronchoscopy is one of the most important diagnostic tools to show the localization and severity of the compression and the airway malacia;[1] however, it requires experience, anesthesia, and can be performed in only a small number of centers in our country.[3,4]

Given the association of chronic airway compression with morbidity and mortality in children, thorough preoperative anatomical and functional assessment is essential. Decisions regarding surgical or conservative management should be individualized based on these evaluations.

Despite the relatively low prevalence of airway compressions in children, increasing awareness and understanding of this condition are essential. However, data on cardiovascular system-related airway compressions in children remain limited. In this study, we aim to evaluate the clinical characteristics and short-term outcomes of children with cardiovascular system-related tracheobronchial compression treated at our tertiary center. Our focus on short-term outcomes reflects an effort to better understand immediate postoperative and management challenges, providing insights that may inform clinical decision-making and improve patient care.

MATERIALS and METHODS

We retrospectively evaluated the clinical records of patients aged 0–18 years who were followed up with a diagnosis of airway compression due to cardiovascular diseases between July 2022 and August 2024. All patients were evaluated by the pediatric pulmonology department. CT images were read and reported by the same experienced pediatric radiologist. Airway compressions were classified into two main groups: vascular ring/pulmonary sling (group A) and malposition or enlargement of cardiovascular structures due to cardiac diseases (group B). We included all patients with tracheobronchial compression due to cardiovascular reasons in our study without a threshold.

Patients older than 18 years and those with airway compression due to non-cardiovascular pathologies were excluded. Demographic and clinical characteristics, including age, symptoms, comorbidities, diagnostic procedures, airway compression site, and the cardiovascular structure causing compression, as well as accompanying bronchoscopic findings (if performed), were recorded. Bronchoscopy was performed in cases where clinical findings were atypical or inconclusive to confirm the presence of compression and/or determine its severity. Flexible bronchoscopy was performed either at the bedside in the intensive care unit or in the operating room by a pediatric pulmonologist. Rigid bronchoscopy, on the other hand, was performed in the operating room by a pediatric surgeon. All patients were evaluated preoperatively, and none of the patients developed tracheobronchial compression post-surgery in our study. Management of the compression (surgical/conservative), duration of hospitalization, the need for respiratory support, and short-term clinical outcomes, including mortality and morbidity, were recorded.

This study was approved by the Başakşehir Çam and Sakura City Hospital Ethical Committee of our hospital (No. 2024-148). Informed consent was obtained from the parents of the patients. The study was conducted in accordance with the Declaration of Helsinki.

Statistical Analysis

Statistical analyses were performed using the IBM Statistical Package for the Social Sciences Version 20 for Windows. Continuous variables that were normally distributed were

presented as means and standard deviations, whereas data with asymmetrical distribution were presented as medians and percentiles. Categorical variables were presented as proportions. The Mann–Whitney U test was performed to compare two independent nonparametric variables. Categorical data were analyzed via χ^2 or Fisher's exact tests. A p-value ≤0.05 was considered significant.

RESULTS

The study included 22 patients with a median age of 24 months (IQR 11–29 months); 68.2% (n=15) of them were male. The median age at diagnosis was 15 days (IQR 0–18 months). Twenty of the patients (90.9%) were symptomatic, while only two patients (9.1%) were asymptomatic at the time of diagnosis. Recurrent wheezing and dyspnea were the most common symptoms, each with a frequency of 59.1%. Stridor was present in only 18.2% (n=4) of the patients. Failure to thrive was observed in 59.1% (n=13) of the patients. Comorbidities were present in 50% of the patients. Down syndrome (13.6%, n=3) and DiGeorge syndrome (13.6%, n=3) were present in 6 patients (27.2%). Table 1 shows the baseline demographic and clinical characteristics of the patients.

There were 10 patients (45.5%) in the vascular ring/pulmonary sling group (group A), while 12 patients (54.5%) were in the abnormally enlarged or malpositioned cardiovascular structure due to the underlying congenital heart disease group (group B). Table 2 shows the detailed analysis of cardiovascular lesions and airway compression sites of the patients. The trachea was the most frequently compressed airway structure, with a frequency of 68.2% in total (15 of 22 patients), while the left main bronchus followed with a frequency of 40.9% (9 of 22 patients). Right arcus aorta with an aberrant left subclavian artery and Kommerell's diverticulum was the most frequent vascular ring, seen in 4 of 10 cases (40%) in group A. In group A, the most common affected site was the trachea, with a frequency of 100%, while it was the left main bronchus in group B, which was affected in 8 of 12 patients (66.6%). The pulmonary artery and/or aorta was the most common structure responsible for airway compression in group B (9 of 12 patients, 75%).

Flexible/semirigid bronchoscopy was performed in 7 patients (31.8%), 5 of whom were in group A. The primary diagnosis was made by multidetector computerized tomography in half of the patients (n=11, 50%), while the other half were diagnosed by echocardiography. The frequency of tracheomalacia and bronchomalacia in patients who underwent bronchoscopy was 71.4% (5 of 7) and 28.6% (2 of 7), respectively, while tracheal stenosis was present in 28.6% (2 of 7). Atelectasis was the most common radiological finding, with a frequency of 45.5% (n=10) of the patients, while air-trapping followed (22.7%, n=5).

Table 1. Baseline demographic and clinical characteristics of the patients (n=22)

	n	%
Age, months, median (IQR)	24 (11–29)	
Age at dignosis, months, median (IQR)	0.5 (0–18)	
Sex		
Female	7	31.8
Male	15	68.2
Prematurity	9	40.9
Comorbidities	11	50
Down syndrome	3	13.6
Digeorge syndrome	3	13.6
Cat-eye syndrome	1	4.5
Esophageal atresia	2	9
Situs inversus totalis	1	4.5
Omphalocele	1	4.5
Symptomatic	20	90.9
Asymptomatic	2	9.1
Cough	10	45.5
Wheezing	13	59.1
Stridor	4	18.2
Dyspnea	13	59.1
Extubation failure	3	13.6
Rcurrent pulmonary infections	12	54.5
Dysphagia	2	9.1
Failure to thrive	13	59.1

IQR: Interquartile range

Two patients, aged 8 and 15 years, did not undergo surgery. Both were asymptomatic with mild compression and were diagnosed incidentally. Each had a right aortic arch with an aberrant left subclavian artery and Kommerell's diverticulum. The median age at surgical intervention was 5.5 months (IQR 2.25–9 months). Seven patients (31.8%) required noninvasive ventilation support to discharge home, while one patient required tracheostomy. During the follow-up period, one patient with atrial septal defect/ventricular septal defect/interrupted aorta is still on invasive ventilation with a tracheostomy, while another patient with total anomalous pulmonary venous drainage/double outlet right ventricle/D-malposition is still on noninvasive ventilation support for 16 hours/day. One patient with atrial septal defect/patent ductus arteriosus and right cardiac failure died during follow-up.

Table 2. Cardiovascular lesions and airway compressions

Cardiovascular lesions (n=22)	n	%	Airway compression		
			Trachea	RMB	LMB
			n	n	n
Group A (vasculary ring/pulmonary sling)	10	45.5	10	2	1
RAA with aberrant LSCA with Kommerell's diverticulum	4		4	1	–
Double aortic arch	3		3	–	–
Pulmonary sling	2		2	1	1
Innominate arter anomaly	1		1	–	–
Group B (abnormally enlarged or malposition of the cardiovascular structure due to underlying CHD)					
Compression from pulmonary artery/aorta	12	54.5			
TOF	2		1	–	2
VSD/ASD/PA	1		–	–	1
VSD/ASD/PS	1		–	–	1
Truncus arteriosus/VSD/ASD/RAA	1		1	1	–
ASD/PS/RAA	1		–	1	–
ASD/VSD/interrupted aorta	1		1	1	–
TAPVD/DORV/D-malposition	1		1	–	1
AVSD/PH	1		1	1	–
Compression from cardiac enlargement					
TOF	2		–	–	2
ASD/PDA/rightcardiacfailure	1		–	1	1
Total	22		15	7	9

RMB: Right main bronchus; LMB: Left main bronchus; RAA: Right aortic arch; LSCA: Left subclavian artery; CHD: Congenital heart disease; TOF: Tetralogy of fallot; VSD: Ventricular septal defect; ASD: Atrial septal defect; PA: Pulmonary atresia; PS: Pulmonary stenosis; TAPVD: Total anomalous pulmonary venous drainage; DORV: Double outlet right ventricle; PDA: Patent ductus arteriosus; AVSD: Atrioventricular septal defect; PH: Pulmonary hypertension

Table 3 shows the comparison between groups A and B. The median age at diagnosis was significantly lower in group B. The frequency of stridor was significantly higher in group A. There was no significant difference in the frequency of any other symptoms between the groups. The median total duration (days) of ventilation support was significantly higher in group B. The overall mortality rate was 1 of 22 (4.5%). There was no operation-related morbidity, while only one patient in group B died after discharge due to underlying heart failure.

DISCUSSION

The present study demonstrates the clinical characteristics and short-term clinical outcomes of patients with airway compressions due to cardiovascular causes, which is a relatively rare finding in children. In our study, the trachea was the most frequently compressed area (68.2% of patients), while the left main bronchus followed (40.9%). Symptoms were mostly nonspecific; dyspnea, wheezing, and failure to thrive were the most common symptoms, highlighting the importance of a high index of suspicion for diagnosis. Cardiovascular and pulmonary pathophysiologies are highly correlated; therefore, the diagnosis and management of vascular airway compressions can be challenging for clinicians. Due to nonspecific symptoms and diagnostic challenges, underdiagnosis remains a significant problem.

The median age at diagnosis was 15 days in our study, while in the vascular ring/pulmonary sling group, the median age at diagnosis was 19 months. Similar to our study, Yubbu et al.[2] reported that the mean age at diagnosis in patients with the vascular ring/pulmonary artery sling group (n=45) was 16.8 months, while it was 3 months in patients with congenital heart disease-associated cardiovascular structure-related airway compression (n=28) in their retrospective study over a ten-year period. In our study, 54.5% of the patients were in the abnormally enlarged or malpositioned cardiovascular structure group due to underlying congenital heart

Table 3. Comparison of clinical characteristics between groups

	Group A (n=10)		Group B (n=12)		p
	n	%	n	%	
Age, months, median (IQR)	26.50 (12–108)		23 (10–26)		0.186
Age at diagnosis, months, median (IQR)	19 (4–98)		0 (0–0.50)		0.002
Sex					
Male	8	80	7	58.3	0.381
Comorbidity	3	30	8	66.7	0.198
Cough	5	50	5	41.7	1.00
Wheezing	5	50	8	66.7	0.666
Stridor	4	40	0	0	0.029
Dyspnea	6	60	7	58.3	1.00
Extubation failure	1	10	2	16.7	1.00
Rcurrent pulmonary infections	6	60	6	50	0.691
Failure to thrive	4	40	9	75	0.192
Median age at surgical intervention, months, median (IQR)	15.50 (2.50–57)		4 (2.25–6)		0.129
Duration of ventilation support after surgery, days, median (IQR)	1.50 (0–8.50)		20 (5–35)		0.013
Duration of hospitalisation after surgery, median (IQR)	12.50 (7–25)		40 (25.50–52.50)		0.070

IQR: Interquartile range

disease (group B), which is associated with early diagnosis due to improving antenatal care and the serious course of the disease. However, patients with vascular ring/pulmonary sling often present with milder, nonspecific respiratory symptoms. As the frequency is relatively low, clinicians may not consider a vascular ring diagnosis until the symptoms are unresponsive to medical management. Consequently, delays in diagnosing vascular rings are common.[2,7]

Similar to previous studies, comorbid diseases and genetic syndromes were present in 50% of the patients, with a higher prevalence in group B (66.7%). DiGeorge syndrome and Down syndrome were present in 6 of 22 cases, which were associated with arch anomalies and congenital heart diseases.[8,9]

Stridor, wheezing, recurrent respiratory infections, apneas, persistent atelectasis, and air trapping may be warning signs of airway compression in children with congenital heart disease. Stridor was present in only 4 of 10 patients in the vascular ring/pulmonary artery sling group, while none of the patients experienced life-threatening complications such as respiratory arrest or apnea. Similarly, Yubbu et al.[2] reported that only half of the patients in the vascular ring/pulmonary sling group had stridor, and none had apnea. Suh et al.[7] reported a median age at diagnosis of 7 months, with 31.4% of patients being asymptomatic and only 8.6% having life-threatening complications at the time of diagnosis in their study of 35 patients with vascular rings. Nonspecific symptoms and the lack of life-threatening symptoms may explain the relatively late age at diagnosis in our study.

The most common underlying etiology for airway compression was the abnormally enlarged or malpositioned cardiovascular structure due to underlying congenital heart disease (group B), a heterogeneous group with a frequency of 54.5% in our study. Tracheobronchial compression in complex congenital heart diseases is clinically significant, as it often complicates respiratory management and may lead to prolonged ventilatory support or recurrent respiratory infections. The intricate anatomical relationships between the tracheobronchial tree and cardiovascular structures necessitate a high index of suspicion and detailed diagnostic workup for timely identification. Early recognition and intervention can significantly impact the prognosis and quality of life for affected patients.

Multiple studies have reported that airway compression due to cardiovascular diseases mainly occurs due to underlying vascular rings or pulmonary slings.[2,10] Our center is one of the largest congenital heart disease centers in Turkey, which may explain the relatively high ratio of congenital heart disease-associated airway compression in our study. Similar to

previous studies, the left main bronchus was the most frequently compressed airway structure in group B (8 of 12 patients). Lai et al.[1] also reported that the left main bronchus was the most commonly affected site in 13 of 20 patients with congenital heart disease (hemodynamics associated), with the left pulmonary artery being the most frequently responsible cardiovascular structure (6 of 13 cases). The left main bronchus is surrounded by pulmonary arteries, the ascending aorta, and the left atrium anteriorly, while the descending aorta crosses posteriorly. This close relationship makes the left main bronchus vulnerable to compression if any of these structures are malpositioned or enlarged.[2]

Vascular rings are rare congenital abnormalities, accounting for 1–3% of all congenital cardiac anomalies, and result from embryological derangement of the aortic arches or pulmonary arteries.[7] There were 10 patients (45.5%) in the vascular ring/pulmonary sling group in our study. A right arcus aorta with an aberrant left subclavian artery and Kommerell's diverticulum was the most frequent vascular ring (40%), followed by double aortic arch (30%). While some studies reported that the double aortic arch was the most common vascular ring,[7,11,12] others have found that a right aortic arch with a left-sided patent ductus or a ductus ligamentum was the most common type.[13,14] Yubbu et al.[2] reported that a right arcus aorta with an aberrant left subclavian artery and Kommerell's diverticulum was the most common vascular ring, with a frequency of 18.1%, in their study of 45 patients with vascular rings/pulmonary slings.

Tracheal compression was present in all cases, and all four patients with stridor were in group A in our study. Similar to previous studies, tracheomalacia (n=3) and tracheal stenosis (n=2) were observed in five patients who underwent bronchoscopy.[2] Prolonged compression from the antenatal period and cartilage destruction are reported to cause tracheobronchomalacia in vascular ring patients. Furthermore, tracheomalacia is reported to be the main pathophysiology, rather than extrinsic compression, in patients with innominate artery anomalies, especially in children with esophageal atresia.[3] Consistent with previous literature, there was one patient with an innominate artery anomaly who had tracheal stenosis and tracheomalacia, with accompanying esophageal atresia in our study.

Another patient with tracheal stenosis had a pulmonary sling, which has been reported in up to 70% of patients with pulmonary slings. Yong et al.[15] reported that 63.6% (21 of 33) of patients with pulmonary slings required tracheal surgery, with slide tracheoplasty performed in 52.4% (11 of 21) of those patients. Even though the number of patients with pulmonary slings is small, one of our patients with a pulmonary sling also underwent concomitant tracheoplasty during pulmonary artery sling surgery.

In our study, all patients except for two with a right arcus aorta with an aberrant left subclavian artery, who were diagnosed at 15 years and 8 years old, respectively, underwent surgery. One of them was asymptomatic and diagnosed incidentally, while the other patient had symptoms of asthma and was diagnosed during further evaluation incidentally. However, as the symptoms were controlled with asthma medications and the compression was mild, she was managed conservatively. A systematic review by Biermann et al.[16] also reported that conservative management might be reasonable in asymptomatic or mildly symptomatic cases with a right aortic arch forming a true vascular ring. However, once moderate-to-severe symptoms develop, surgical intervention is definitely indicated.

Even though there is no standard definition for timing and indications for surgery, most studies report that >50% reduction in tracheal airway size and persistent symptoms that affect a patient's quality of life, regardless of the degree of airway compression, are accepted as criteria for surgical intervention.[17] Some studies reported successful outcomes in both symptomatic and asymptomatic patients; however, surgery in asymptomatic cases remains controversial.[18] Present studies report the ratio of surgical intervention for vascular compressions as 52–100%.[7,13,17]

The median age at surgery in group A in our study was 15.50 months (IQR 2.50–57), which was older than in previous studies,[2,14,19] likely related to an older age at diagnosis. Evans et al.[14] reported that the age at surgery for vascular rings has decreased over the past 25 years with improving prenatal diagnosis. While the median ventilation support duration after surgery was 1.5 days, the median discharge day was 12 days in our study, which was lower than in group B. There was no surgery-related mortality in either group. Suh et al.[7] reported that the median hospital stay was 17 days, while the median duration in the intensive care unit was 3.5 days in their study, which included vascular ring patients. As group B had more complex cardiac diseases, longer hospital stays in group B were unsurprising. Similar to previous studies, the short-term prognosis of vascular rings was good.

The overall mortality rate was 1 in 22 (4.5%) patients, who was in group B and died after hospital discharge due to underlying heart failure. Accompanying complex heart disease is reported as the only factor affecting the mortality rate in vascular ring patients.[7]

There are some limitations to our study. First, the study was designed as a retrospective study. In addition, as our study included a two-year period at a single center, the sample size is relatively small. Long-term follow-up of the patients is required to understand the long-term clinical outcomes of these patients. Despite these limitations, our study includes a wide variety of cardiovascular causes of tracheobronchial compressions in children.

CONCLUSION

In conclusion, tracheobronchial compressions are important comorbidities in children with cardiovascular diseases that may complicate disease management. As the symptoms are mostly nonspecific, the possibility of vascular compression should be considered for patients with an atypical clinical course. Comorbidities, underlying cardiac disease, and the severity of airway compression can affect morbidity and mortality rates. With early diagnosis and timely referral for surgery in symptomatic patients, the prognosis is expected to be good, especially for isolated vascular rings. As there is no standard diagnostic algorithm, the diagnosis and management of this complex group of patients are highly challenging for clinicians. Long-term, large-scale studies are needed in this area to standardize the diagnostic and therapeutic management of these patients.

Disclosures

Ethics Committee Approval: The study was approved by the Başakşehir Çam and Sakura City Hospital Ethics Committee (No: 2024-148, Date: 14/08/2024).

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