

Early outcomes of surgical repair of vascular rings in infants and young children: a retrospective cohort study

Aleksey V. Bedin¹, Yuriy N. Shamrin¹, Mikhail A. Abramyan^{1,2}, Evgenia S. Korochkina¹, Ksenia A. Khasanova^{1,3}, Maria M. Kurako¹, Anastasiya Yu. Gotskina¹, Vladimir P. Miroshnichenko^{1,2}, Dmitriy Yu. Ovsyannikov²

Corresponding author: Aleksey V. Bedin,
alekseybedin@gmail.com

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Informed consent

The patient's official representative has provided informed consent for the use of the recordings for medical purposes.

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Conflict of interest

The authors declare no conflict of interest.

Ethics approval

This retrospective study was approved by the local ethics committee of Morozov Children's City Clinical Hospital (Protocol No. 7 dated December 10, 2025). The study was conducted in accordance with the ethical principles of the World Medical Association Declaration of Helsinki, as amended in October 2024. Consent was obtained from the patients' legal representatives by telephone. Patient data were anonymized before analysis.

Contribution of the authors

Concept and study design: A.V. Bedin

Data collection and analysis: A.V. Bedin, Yu.N. Shamrin

Statistical analysis: A.V. Bedin

Drafting the article: A.V. Bedin, Yu.N. Shamrin, A.Yu. Gotskina, E.S. Korochkina

Critical revision of the article: M.M. Kurako, M.A. Abramyan, K.A. Khasanova

Final approval of the version to be published: all authors

ORCID

A.V. Bedin, <https://orcid.org/0000-0001-8489-6438>

Yu.N. Shamrin,
<https://orcid.org/0000-0002-2260-9533>

M.A. Abramyan,
<https://orcid.org/0000-0003-4018-6287>

E.S. Korochkina,
<https://orcid.org/0000-0003-3537-6262>

K.A. Khasanova,
<https://orcid.org/0000-0001-6926-3165>

¹ Morozov Children's City Clinical Hospital, Moscow, Russian Federation

² Patrice Lumumba Peoples' Friendship University of Russia, Moscow, Russian Federation

³ Sechenov First Moscow State Medical University (Sechenov University), Moscow, Russian Federation

Abstract

Introduction: Vascular compression of the tracheobronchial tree (TBT) and esophagus occurs in congenital anomalies of the aortic arch, its branches, and the pulmonary arteries. Although the vast majority of patients with this condition are symptomatic, vascular compression is often not diagnosed promptly. The clinical presentation results from compression of the trachea and/or esophagus; therefore, the predominant manifestations are dyspnea (stridor, cough, and shortness of breath) and/or dysphagia (choking, difficulty swallowing, and regurgitation). Diagnostic approaches and methods of surgical treatment remain clinically relevant. The timing of and indications for surgical correction remain controversial, and outcome data are limited to a small number of publications.

Objective: To evaluate the effect of surgical treatment of vascular rings on symptom resolution during the early postoperative period in infants and young children; to define indications for surgical treatment; to report the frequency of aortopexy used to prevent residual symptoms according to vascular ring anatomy; and to assess the incidence of chylothorax and its association with the anatomical type of vascular ring.

Methods: This retrospective, nonrandomized cohort study included 58 pediatric patients who underwent surgery for an isolated congenital vascular ring causing compression of the TBT and esophagus at a single center between January 2018 and July 2024. Patients were divided into four groups according to vascular ring anatomy: double aortic arch ($n = 16$; 27.6%), right aortic arch with a left ligamentum arteriosum ($n = 34$; 58.6%), left aortic arch with a right ligamentum arteriosum ($n = 4$; 6.9%), and pulmonary artery sling ($n = 4$; 6.9%). All patients underwent contrast-enhanced multidetector computed tomography for diagnostic confirmation and surgical planning. Early symptom resolution, technical aspects of surgical correction, postoperative length of stay, and postoperative complications were assessed.

Results: The median age at surgery was 7 months (Q1–Q3, 4–25 months). Preoperative symptoms (dyspnea and/or dysphagia) were present in 84.5% of patients. There were no statistically significant between-group differences in anthropometric characteristics, age, or symptom status ($p > 0.05$). Complete symptom resolution by hospital discharge was achieved in 83.7% of symptomatic patients. Length of hospital stay differed significantly among the groups ($p = 0.028$), with the highest median in the pulmonary artery sling group (18 hospital days), owing to the need for cardiopulmonary bypass and a more severe baseline condition. Chylothorax was the most common complication, occurring in 13 patients (22.4%) and most frequently in those with a double aortic arch (43.8%). Aortopexy was performed most frequently in patients with a right aortic arch and left ligamentum arteriosum (70.6%; $p = 0.001$). There were no deaths.

Conclusion: Surgical correction of vascular rings in infants and young children was highly effective in the early postoperative period, with complete symptom resolution in 83.7% of symptomatic patients and no mortality. These findings support the safety of surgery when combined with early diagnosis and a multidisciplinary approach. In our opinion, for a vascular ring formed by a right aortic arch and left ligamentum arteriosum, adding aortopexy to division of the ligamentum arteriosum may visibly eliminate residual esophageal compression. The long-term clinical benefit of this approach requires further evaluation. The principal clinical concern is the substantial incidence of postoperative

M.M. Kurako,
<https://orcid.org/0000-0002-1100-1506>

A.Yu. Gotskina, <https://orcid.org/0009-0004-8635-3663>

V.P. Miroshnichenko,
<https://orcid.org/0000-0003-4140-8777>

D.Yu. Ovsyannikov, <https://orcid.org/0000-0002-4961-384X>

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chylothorax (22.4%), underscoring the need to improve surgical techniques and management protocols and to identify the underlying causes.

Keywords: aortopexy; chylothorax; esophageal compression; Kommerell diverticulum; respiratory disorders; surgical treatment; tracheal compression; tracheobronchomalacia; vascular ring

Непосредственные результаты хирургического лечения сосудистых колец у младенцев и детей раннего возраста: ретроспективное когортное исследование

А.В. Бедин¹, Ю.Н. Шамрин¹, М.А. Абрамян^{1,2}, Е.С. Корочкина¹, К.А. Хасанова^{1,3}, М.М. Курако¹, А.Ю. Гоцкина¹, В.П. Мирошник^{1,2}, Д.Ю. Овсянников²

Для корреспонденции: Алексей Владимирович Бедин, alekseybedin@gmail.com

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Информированное согласие

Получено информированное согласие официальных представителей пациентов на публикацию и использование медицинских данных в научных целях.

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¹ Государственное бюджетное учреждение здравоохранения города Москвы «Морозовская детская городская клиническая больница Департамента здравоохранения города Москвы», Москва, Российская Федерация

² Федеральное государственное автономное образовательное учреждение высшего образования «Российский университет дружбы народов имени Патриса Лумумбы», Москва, Российская Федерация

³ Федеральное государственное автономное образовательное учреждение высшего образования Первый Московский государственный медицинский университет имени И.М. Сеченова Министерства здравоохранения Российской Федерации (Сеченовский Университет), Москва, Российская Федерация

Аннотация

Актуальность. Сосудистая компрессия трахеобронхиального дерева (ТБД) и пищевода возникает при врожденных аномалиях развития дуги аорты, ее ветвей и легочных артерий. Несмотря на то, что симптомы наблюдаются у подавляющего большинства пациентов с этой патологией, диагноз сосудистой компрессии зачастую не подтверждается своевременно. Клиническая картина патологии обусловлена сдавлением трахеи и/или пищевода, и, таким образом, ведущими симптомами будут диспноэ (стридорозное дыхание, кашель, одышка) и/или дисфагия (поперхивание, затруднение глотания, срыгивание). Актуальными остаются подходы к диагностике порока и способам хирургического лечения. Вопросы сроков и показаний к хирургической коррекции данной патологии остаются дискуссионными, а результаты представлены единичными публикациями.

Цель. Оценить эффект хирургического лечения сосудистых колец на регресс симптоматики в раннем послеоперационном периоде у младенцев и детей раннего возраста. Определить показания к проведению хирургического лечения сосудистых колец. Представить частоту применения аортотомии для профилактики резидуальных симптомов в зависимости от типа сосудистых колец. Оценить частоту развития хилоторакса и зависимость от анатомического типа сосудистых колец.

Методы. Выполнено ретроспективное когортное нерандомизированное исследование, основанное на анализе 58 историй болезни пациентов детского возраста, оперированных по поводу врожденного порока сердца, представленного изолированным сосудистым кольцом с компрессией ТБД и пищевода, в одном центре за период с января 2018 г. по июль 2024 г. В зависимости от анатомического типа порока пациенты были разделены на четыре группы: двойная дуга аорты ($n = 16$; 27,6%), правая

Вклад авторов

Концепция и дизайн работы: А.В. Бедин
Сбор и анализ данных: А.В. Бедин, Ю.Н. Шамрин
Статистическая обработка данных: А.В. Бедин
Написание статьи: А.В. Бедин, Ю.Н. Шамрин, А.Ю. Гоцкина, Е.С. Корочкина
Исправление статьи: М.М. Курако, М.А. Абрамян, К.А. Хасанова
Утверждение окончательного варианта статьи: все авторы

ORCID

А.В. Бедин,
<https://orcid.org/0000-0001-8489-6438>
Ю.Н. Шамрин,
<https://orcid.org/0000-0002-2260-9533>
М.А. Абрамян,
<https://orcid.org/0000-0003-4018-6287>
Е.С. Корочкина,
<https://orcid.org/0000-0003-3537-6262>
К.А. Хасанова,
<https://orcid.org/0000-0001-6926-3165>
М.М. Курако,
<https://orcid.org/0000-0002-1100-1506>
А.Ю. Гоцкина,
<https://orcid.org/0009-0004-8635-3663>
В.П. Мирошниченко,
<https://orcid.org/0000-0003-4140-8777>
Д.Ю. Овсянников,
<https://orcid.org/0000-0002-4961-384X>

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дуга аорты с левой артериальной связкой ($n = 34$; 58,6%), левая дуга аорты с правой артериальной связкой ($n = 4$; 6,9 %) и слинг легочной артерии ($n = 4$; 6,9 %). Всем пациентам выполнялась мультиспиральная компьютерная томография с контрастным усилением для верификации диагноза и планирования операции. Оценивались непосредственная эффективность операции в отношении устранения симптоматики, технические аспекты хирургической коррекции, длительность послеоперационного периода и анализ осложнений коррекции.

Результаты. Медиана возраста на момент операции составила 7 месяцев (Q1–Q3: 4–25 месяцев). Симптомы (диспноэ и/или дисфагия) наблюдались у 84,5 % пациентов до операции. Статистически значимых различий по антропометрическим данным, возрасту и наличию симптомов между группами не выявлено ($p > 0,05$). Полный регресс симптоматики к моменту выписки достигнут у 83,7 % симптомных пациентов. Выявлены статистические различия в продолжительности госпитализации между исследуемыми группами ($p = 0,028$) с наибольшей медианой в группе слинга легочной артерии ($Me = 18$ койко-дней), что связано с необходимостью применения искусственного кровообращения во время операции и более тяжелым исходным статусом. Наиболее частым осложнением явился хилоторакс, развившийся у 13 (22,4 %) пациентов и чаще регистрировавшийся при двойной дуге аорты (43,8 %). Аортотомия чаще выполнялась при правой дуге аорты с левой артериальной связкой (70,6 %; $p = 0,001$). Летальных исходов не было.

Заключение. Хирургическая коррекция сосудистых колец у младенцев и детей раннего возраста продемонстрировала в раннем послеоперационном периоде высокую эффективность (полное устранение симптомов у 83,7 % пациентов) и отсутствие летальности, что подтверждает безопасность вмешательства при условии мультидисциплинарного подхода и ранней диагностики. По нашему мнению, при сосудистом кольце, представленном правой дугой аорты с левой артериальной связкой, аортотомия в дополнение к пересечению связки артериального протока способствует визуальному устранению резидуальной компрессии пищевода. Отдаленный клинический эффект этого метода требует дальнейшей оценки. Основной клинической проблемой остается значительная частота послеоперационного хилоторакса (22,4 %), определяющая необходимость дальнейшего совершенствования хирургической техники и протоколов ведения и определения причин его развития.

Ключевые слова: дивертикул Коммереля; дыхательные расстройства; компрессия пищевода; компрессия трахеи; сосудистое кольцо; трахеобронхомалия; хирургическое лечение сосудистых колец



Introduction

Vascular compression of the tracheobronchial tree (TBT) and esophagus occurs in congenital anomalies of the aortic arch, its branches, and the pulmonary arteries. This condition is relatively rare and accounts for 0.5–1.0% of all congenital cardiovascular anomalies [1; 2]. Hommel provided the first description of this vascular anomaly in 1737, while the term “vascular ring” was first introduced by R. Gross in 1945 in connection with surgical correction of a double aortic arch [3]. Anatomically, vascular rings are classified as complete or incomplete. Complete rings are formed by a double aortic arch; a right aortic arch with a left patent ductus arteriosus (PDA) or ligamentum arteriosum; or a right aortic arch with an aberrant right subclavian artery and a left PDA. Incomplete vascular

rings (vascular slings) are formed by an aberrant right subclavian artery (*arteria lusoria*) or a left pulmonary artery arising anomalously from the right pulmonary artery (pulmonary artery sling) [4; 5].

The clinical presentation is caused by compression of the trachea and/or esophagus; therefore, the predominant manifestations are dyspnea (stridor, cough, and shortness of breath) and/or dysphagia (choking, difficulty swallowing, and regurgitation). These symptoms are entirely nonspecific, and differential diagnosis must include upper airway disorders, bronchopulmonary disease, and gastrointestinal disease [6; 7]. In addition to overt clinical manifestations, patients are at high risk of tracheobronchomalacia, which may be asymptomatic [8]. Importantly, the absence of symptoms does not rule

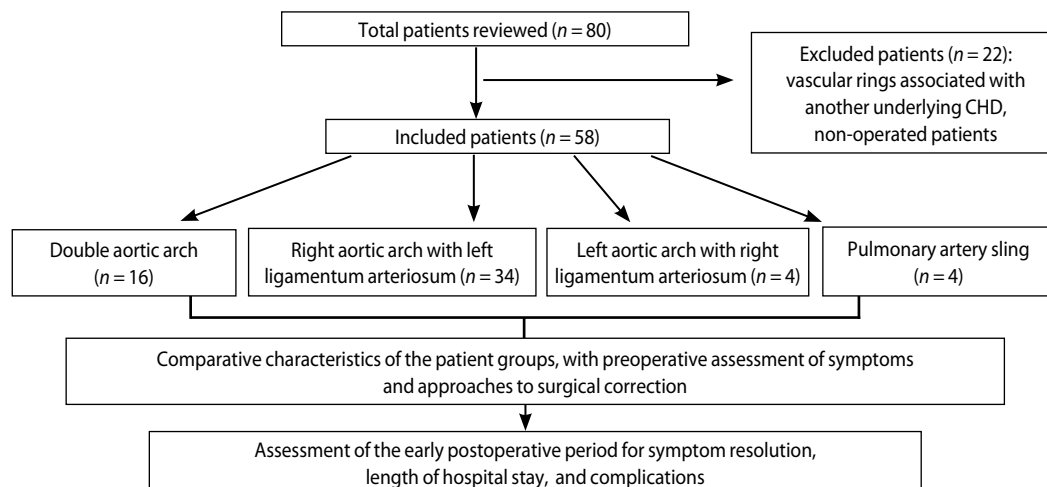


Fig. 1. Study flowchart

Note. CHD, congenital heart disease.

out substantial tracheal compression. The anatomical effects of a vascular ring may be underestimated when clinical decisions are based solely on symptoms of tracheal compression. Routine studies such as chest radiography and echocardiography do not always identify this condition, although echocardiography is highly specific in prenatal diagnosis. Gastroscopy, bronchoscopy, and contrast esophagography may demonstrate the abnormality without establishing its cause. Contrast-enhanced multidetector computed tomography (MDCT) is the most accurate diagnostic modality, reliably defining the anatomical type of anomaly and the location and severity of TBT compression [9].

When surgery is performed promptly, most patients experience complete symptom resolution and improved quality of life [10; 11].

Russian-language literature contains very few studies of vascular anomalies causing compression of the TBT and esophagus. Available publications are largely limited to small case series, individual case reports, or the experience of thoracic surgeons. The present study summarizes a single center's experience with a comprehensive approach to diagnosis, surgical treatment, and postoperative management of patients with this rare and previously unrecognized condition.

The objectives of this study were to evaluate the effect of surgical treatment of vascular rings on symptom resolution during the early postoperative period in infants and young children; define indications for surgery; assess the frequency of aortopexy performed to prevent residual

symptoms according to vascular ring anatomy; and determine the incidence of chylothorax and its association with the anatomical type of vascular ring.

Methods

Study design

This was a retrospective, nonrandomized cohort study. A total of 80 medical records of patients with vascular anomalies causing tracheal and esophageal compression were reviewed.

Inclusion criteria

The study included 58 consecutive pediatric patients who underwent surgery at a single hospital between January 2018 and July 2024 for one of four anatomical types of vascular ring: double aortic arch, right aortic arch with a left ligamentum arteriosum (hereinafter, right aortic arch with left ligamentum), left aortic arch with a right ligamentum arteriosum (hereinafter, left aortic arch with right ligamentum), or pulmonary artery sling.

Exclusion criteria

Patients with a diagnosed vascular ring who did not undergo surgery were excluded, as were patients with a vascular ring associated with another congenital heart defect requiring correction; anomalies in the position or origin of the brachiocephalic vessels causing TBT and esophageal compression (*arteria lusoria*); or isolated crossing of the pulmonary artery branches causing tracheal compression (Fig. 1).

Study hypothesis and endpoints

We hypothesized that surgical correction of a congenital great-vessel anomaly forming a vascular ring and compressing the intrathoracic structures enclosed within it (the TBT and esophagus) would completely eliminate respiratory and gastrointestinal symptoms. In asymptomatic patients with compression confirmed by MDCT angiography, surgery was expected to prevent the development of tracheobronchomalacia.

The prespecified study endpoints were the number of patients in whom respiratory and gastrointestinal symptoms resolved, the number of early surgical complications, and the effects of these complications on length of hospital stay.

Diagnostic algorithm and indications for surgery

The diagnostic algorithm included obtaining a medical history and interviewing the patients' parents to determine the frequency and severity of respiratory symptoms (dyspnea), including stridor, cough, and a history of acute respiratory viral infections, bronchitis, or pneumonia, as well as swallowing disorders (dysphagia). Based on these data, patients were classified as symptomatic or asymptomatic. Echocardiography was the initial diagnostic modality and was used to identify the dominant aortic arch and branching pattern of the brachiocephalic vessels. When echocardiographic findings suggested a vascular ring, all patients underwent MDCT with three-dimensional reconstruction to assess the spatial relationships between the vessels and thoracic organs, including the trachea, bronchi, and esophageal course.

Patients were classified as symptomatic if the "Presenting complaints" or "History of present illness" sections of the medical record documented manifestations characteristic of respiratory dysfunction (cough, choking, or stridor) and/or gastrointestinal symptoms (difficulty swallowing solid food or regurgitation). When a vascular ring was confirmed by imaging and relevant specialists had excluded primary respiratory or gastrointestinal disease, these symptoms were considered indications for surgery. In asymptomatic patients, surgery was indicated when a vascular ring was accompanied by visualized tracheal deviation or stenosis of the trachea and/or main bronchus, defined as luminal narrowing of at least 20%. In symptomatic patients, these anatomical abnormalities were considered additional indications beyond the existing clinical manifestations.

In 8 patients (14%), MDCT showed no visible tracheal or bronchial narrowing despite marked dysphagia. These patients underwent biplanar fluoroscopic

esophagography with passage of a water-soluble iodinated contrast agent. Esophageal narrowing greater than 50% was considered an indication for surgery.

Surgical techniques according to anatomical type

Pulmonary artery sling. Pulmonary artery sling was repaired using the standard technique through a median sternotomy under normothermic cardiopulmonary bypass [12]. The PDA was isolated and divided. The anomalously positioned left pulmonary artery was mobilized along its entire course, divided, brought out from beneath the trachea, and reimplanted into the pulmonary trunk in an orthotopic position.

Double aortic arch. In patients with a double aortic arch, thoracotomy was performed through the third or fourth intercostal space: a left thoracotomy for a dominant right arch and a right thoracotomy for a dominant left arch. The rudimentary arch was isolated and clamped in an area without brachiocephalic branches, then divided; both stumps were closed with two-layer polypropylene sutures. This opened the ring and relieved compression of the TBT and esophagus. Aortopexy was generally unnecessary during double aortic arch repair because no Kommerell diverticulum was present.

Right aortic arch with a left ligamentum arteriosum and left aortic arch with a right ligamentum arteriosum. The descending aorta, ligamentum arteriosum, Kommerell diverticulum, and subclavian artery arising from the diverticulum were exposed in the accessible operative field. The ligamentum arteriosum was divided and oversewn at both ends. When the diverticulum was sufficiently large to compress the esophagus and posed a risk of future aneurysmal dilatation, it was resected and oversewn as close as possible to the descending aorta, followed by reimplantation of the left subclavian artery into the left common carotid artery.

In 50% of cases ($n = 29$), the repair was supplemented with posterior aortopexy to the base of the inner surface of the third rib using pledgeted U-shaped monofilament sutures with polytetrafluoroethylene pledgets. Aortopexy was performed when visual inspection demonstrated residual esophageal compression by the Kommerell diverticulum or when preoperative computed tomography showed a diverticular diameter more than 1.5 times that of the descending aorta. A right thoracotomy was used for a left aortic arch with a right ligamentum arteriosum; sternotomy was used in one case.

In patients with preoperatively confirmed stenosis of the distal trachea and/or bronchi, surgery was performed with intraoperative bronchoscopic guidance ($n = 46$; 79.3%).

Table 1. Distribution of patients according to vascular ring type

Group	<i>n</i>	%	95% CI
1. Double aortic arch	16	27.6	16.7–40.9
2. Right aortic arch with left ligamentum arteriosum	34	58.6	44.9–71.4
3. Left aortic arch with right ligamentum arteriosum	4	6.9	1.9–16.7
4. Pulmonary artery sling	4	6.9	1.9–16.7

Statistical analysis

Statistical analysis was performed using StatTech version 4.6.1 (StatTech LLC, Russia). The Shapiro–Wilk test was used to assess the normality of continuous variables. Non-normally distributed continuous data are presented as the median (Me) and lower and upper quartiles (Q1–Q3). Four-group comparisons of non-normally distributed continuous variables were performed using the Kruskal–Wallis test, followed by Dunn’s *post hoc* test with Holm adjustment. Proportions in 2×2 contingency tables were compared using Fisher’s exact test when expected cell counts were less than 10. Differences were considered statistically significant at $p < 0.05$. Owing to the small

numbers of patients in groups 3 and 4, the between-group analyses had low statistical power. No sample-size calculation was performed because this retrospective study included all consecutive patients who underwent surgery during the specified period.

Results

In all 58 patients, the final clinical diagnosis was established by MDCT. Patients were divided into four groups according to anatomical type. Table 1 presents the distribution of patients by vascular ring type. The most common anatomy was a right aortic arch with a left ligamentum arteriosum or PDA, observed in 58.6% of patients.

Fig. 2. Three-dimensional reconstruction of a double aortic arch (arrows indicate anatomical landmarks):

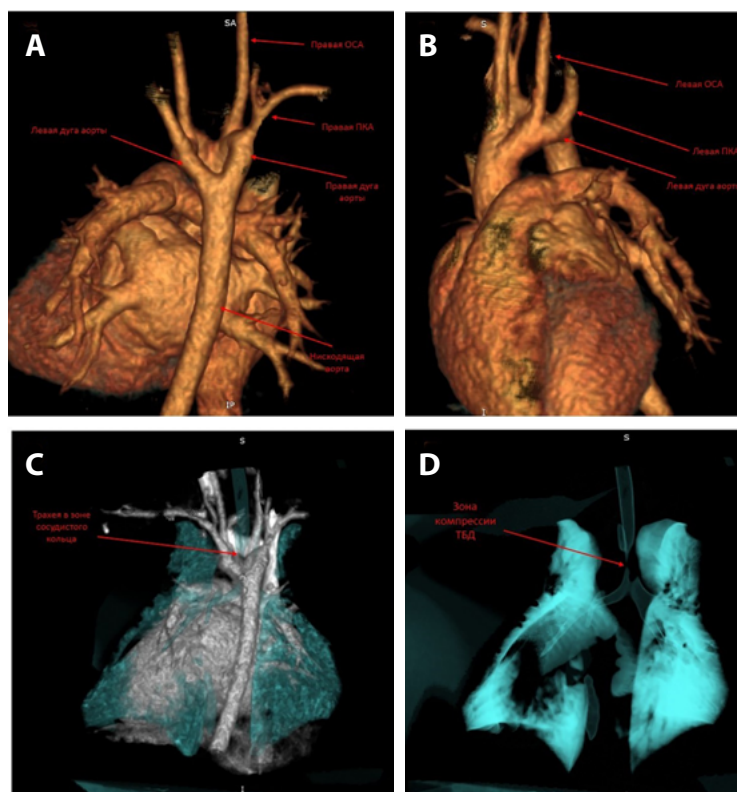
(A) frontal orientation, posterior view, showing the right and left aortic arches, descending aorta, right common carotid artery, and right subclavian artery;

(B) frontal orientation, anterior view, showing the left aortic arch, left subclavian artery, and left common carotid artery;

(C) frontal orientation, posterior view, showing vessels and portions of the lung parenchyma, with the arrow indicating the trachea within the vascular ring;

(D) frontal orientation, posterior view, showing the tracheobronchial tree and lung parenchyma, with the arrow indicating the site of tracheal compression by the vascular ring

Note. TBT, tracheobronchial tree.



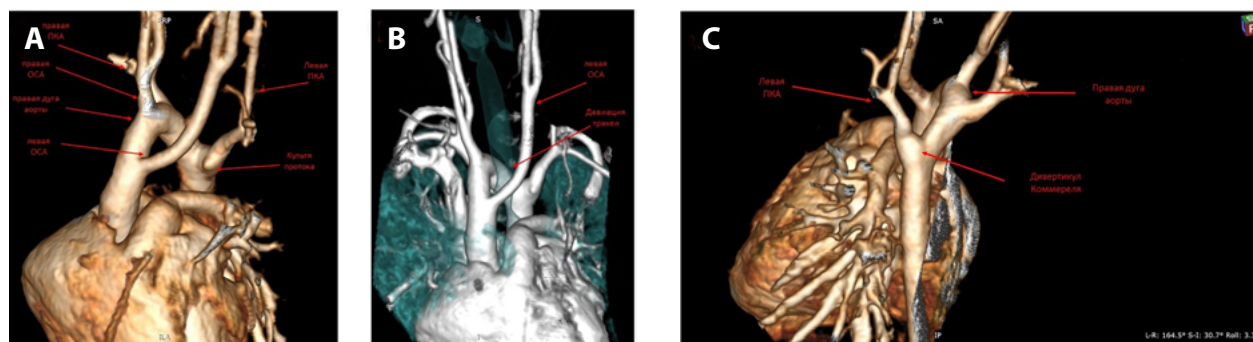


Fig. 3. Three-dimensional reconstruction of a vascular ring formed by a right aortic arch and ligamentum arteriosum (arrows indicate anatomical landmarks): (A) frontal orientation, anterior view, showing the right aortic arch, right common carotid and subclavian arteries, left common carotid and subclavian arteries, and ductal stump; (B) frontal orientation, anterior view, showing vessels and portions of the lung parenchyma, with tracheal deviation and the left common carotid artery indicated; (C) frontal orientation, posterior view, showing the right aortic arch, left subclavian artery, and Kommerell diverticulum

Note. CCA, common carotid artery; SA, subclavian artery.

Figures 2–4 show MDCT-based three-dimensional reconstructions of the vascular ring variants, depicting both the isolated vascular anatomy and its effects on the TBT and esophagus. All images were reconstructed from the patients' original DICOM files using RadiAnt DICOM Viewer 2025.1 (Poland).

A comparative analysis of anthropometric characteristics and age among the groups is presented in Table 2.

The median age was 7 months (Q1–Q3, 4–25 months). The study included 37 boys (63.8%) and 21 girls (36.2%).

The groups were comparable in height and weight, with no statistically significant differences in body surface area or age ($p = 0.447$ and $p = 0.430$, respectively).

The primary outcome of vascular ring repair was the resolution of symptoms associated with TBT and esophageal compression. Before surgery, most patients had clinical manifestations (49 patients; 84.5%; 95% CI, 72.6–92.7%), with no between-group difference in preoperative symptom status ($p = 0.634$). All patients with a left aortic arch and right ligamentum arteriosum or a pulmonary artery sling were symptomatic.

Fig. 4. Left pulmonary artery sling: (A) three-dimensional reconstruction of the heart and great vessels, frontal orientation, posterior view, with the arrow indicating the origin of the aberrant left pulmonary artery and a tube within the esophagus; (B) axial view, with arrows indicating the sites of esophageal and tracheal compression by the left pulmonary artery; (C) frontal orientation, anterior view, showing vessels and portions of the lung parenchyma, with arrows indicating the trachea, esophagus containing a tube, and aberrant left pulmonary artery; (D) frontal orientation, posterior view, showing the tracheobronchial tree and lung parenchyma, with the arrow indicating the site of compression

Note. PA, pulmonary artery; TBT, tracheobronchial tree.

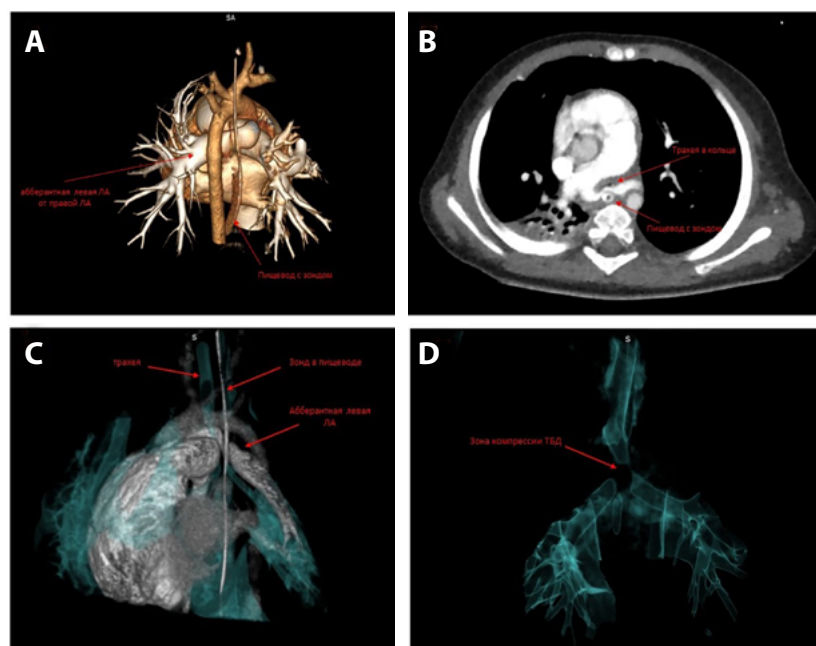


Table 2. Anthropometric characteristics and age according to vascular ring type

Variable	Vascular ring type	Median	Q1–Q3	n	p
Weight, kg	Double aortic arch	7.03	5.60–11.82	16	0.459
	Right aortic arch with left ligamentum arteriosum	7.47	6.54–12.38	34	
	Left aortic arch with right ligamentum arteriosum	11.00	6.99–16.25	4	
	Pulmonary artery sling	6.03	4.33–8.11	4	
Height, cm	Double aortic arch	62.50	59.25–89.75	16	0.253
	Right aortic arch with left ligamentum arteriosum	66.50	62.00–85.50	34	
	Left aortic arch with right ligamentum arteriosum	67.00	65.50–88.50	4	
	Pulmonary artery sling	59.50	55.25–63.75	4	
Body surface area, m ²	Double aortic arch	0.33	0.30–0.57	16	0.447
	Right aortic arch with left ligamentum arteriosum	0.38	0.33–0.55	34	
	Left aortic arch with right ligamentum arteriosum	0.37	0.36–0.57	4	
	Pulmonary artery sling	0.32	0.26–0.38	4	
Age at surgery, months	Double aortic arch	6	4–30	16	0.430
	Right aortic arch with left ligamentum arteriosum	7	4–20	34	
	Left aortic arch with right ligamentum arteriosum	23	9–45	4	
	Pulmonary artery sling	5	2–7	4	

Symptomatic patients also predominated in the double aortic arch and right aortic arch with left ligamentum arteriosum groups, with comparable proportions in these two groups (Table 3).

Respiratory symptoms were more prevalent than swallowing disorders in each group. The distribution of symptoms is shown in Table 4.

Complete symptom resolution by hospital discharge was achieved in 41 symptomatic patients (83.7%). Early postoperative improvement without complete symptom resolution occurred in two children with a double aortic

arch: one had persistent stridor and the other had exertional dyspnea. Two patients with a ring formed by a right aortic arch and left ligamentum arteriosum also improved without complete resolution: one had tachypnea and reduced exercise tolerance, while the other had a pre-existing tracheostomy and was not decannulated during the early postoperative period. Owing to more severe baseline TBT abnormalities, all four patients with a pulmonary artery sling improved without complete resolution of respiratory failure in the early postoperative period. Three were extubated after repair,

Table 3. Preoperative symptom status according to vascular ring type

Status	Double aortic arch, n (%)	Right aortic arch with left ligamentum, n (%)	Left aortic arch with right ligamentum, n (%)	Pulmonary artery sling, n (%)	p
Symptomatic	13 (81.2)	28 (82.4)	4 (100.0)	4 (100.0)	0.634
Asymptomatic	3 (18.8)	6 (17.6)	0 (0.0)	0 (0.0)	

Table 4. Symptoms according to vascular ring type

Symptom	Status	Double aortic arch, <i>n</i> (%)	Right aortic arch with left ligamentum, <i>n</i> (%)	Left aortic arch with right ligamentum, <i>n</i> (%)	Pulmonary artery sling, <i>n</i> (%)	<i>p</i>
Dysphagia	Present	7 (43.8)	14 (41.2)	2 (50.0)	1 (25.0)	0.896
	Absent	9 (56.2)	20 (58.8)	2 (50.0)	3 (75.0)	
Dyspnea	Present	11 (68.8)	20 (58.8)	4 (100.0)	4 (100.0)	0.171
	Absent	5 (31.2)	14 (41.2)	0 (0.0)	0 (0.0)	

while one required slide tracheoplasty during the early postoperative period to restore normal airway patency.

There were no deaths in any group. Figure 5 compares length of hospital stay among vascular ring types. The highest median, 18 hospital days, occurred in the pulmonary artery sling group, reflecting more severe baseline respiratory failure and the use of cardiopulmonary bypass. Length of hospital stay differed significantly among the groups ($p = 0.028$). *Post hoc*

analysis, however, revealed no statistically significant pairwise differences.

During the initial phase of the study (2018–2022), all patients underwent intraoperative tracheoscopy and gastroscopy to assess the need for aortopexy. Subsequently, aortopexy was performed when a Kommerell diverticulum was considered large, defined as a diameter more than 1.5 times that of the descending aorta, and continued to compromise the esophagus

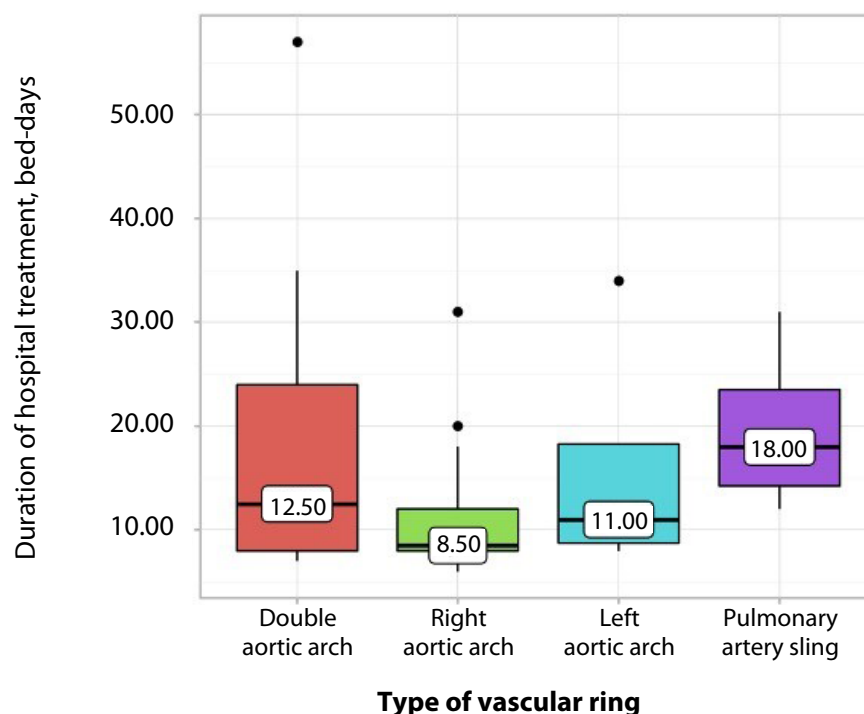


Fig. 5. Length of hospital stay, days. Kruskal – Wallis test, $p = 0.028$

Table 5. Aortopexy according to vascular ring type

Aortopexy	Double aortic arch, n (%)	Right aortic arch with left ligamentum, n (%)	Left aortic arch with right ligamentum, n (%)	Pulmonary artery sling, n (%)	p
Not performed	11 (68.8)	10 (29.4)	4 (100.0)	4 (100.0)	0.001*
Performed	5 (31.2)	24 (70.6)	0 (0.0)	0 (0.0)	

* Statistically significant difference ($p < 0.05$).

and TBT after division of the ligamentum arteriosum. The frequency of aortopexy differed significantly among vascular ring types ($p = 0.001$). Aortopexy was not performed in patients with a left aortic arch and right ligamentum arteriosum or in those with a pulmonary artery sling. It was used most frequently in patients with a right aortic arch and left ligamentum arteriosum (24 patients; 70.6%), owing to the anatomical characteristics of the diverticulum in this group (Table 5).

Chylothorax, a complication characteristic of this repair, occurred frequently during the early postoperative period and became apparent from postoperative day 2 as chylous drainage. This complication required prolonged conservative treatment, including octreotide infusion and temporary discontinuation of enteral feeding, and substantially increased length of hospital stay. Postoperative chylothorax occurred in 13 of all surgically treated patients (22.4%; 95% CI, 10.0–31.9%). The median age of patients with chylothorax was 16 months (Q1–Q3, 5–39 months), and their median weight was 8.6 kg (Q1–Q3, 7.00–15.00 kg). This complication occurred most frequently in patients with a double aortic arch ($n = 7$; 43.8%). Nevertheless, the incidence of chylothorax did not differ significantly among the vascular ring types ($p = 0.089$), likely because of the unequal group sizes (Table 6).

Among the 10 patients whose chylothorax was treated conservatively, the median length of hospital stay was 16 days (Q1–Q3, 12.0–16.5 days). In contrast, among the three patients who required thoroscopic clipping of

the thoracic duct in addition to conservative therapy, the median length of stay was significantly longer at 46 days (Q1–Q3, 38.0–55.5 days).

Intraoperative aortopexy was not associated with the incidence of postoperative chylothorax ($p = 1.000$). The occurrence of this complication was also unrelated to the operating surgeon or differences in surgical approach.

Discussion

A vascular ring is a rare group of congenital anomalies of the intrathoracic great vessels that causes compression of the tracheobronchial tree and esophagus. Advances in modern diagnostic methods have enabled earlier and more accurate detection of vascular TBT compression [1; 13]. Greater awareness among pulmonologists and gastroenterologists of the potential vascular origin of dyspnea and dysphagia promotes earlier recognition of vascular rings and timely surgical correction. At our multidisciplinary children's hospital, the increased detection of this condition resulted from the involvement of pediatric pulmonologists, otorhinolaryngologists, and thoracic surgeons in the diagnostic process and the establishment of an interdisciplinary algorithm. Most patients were referred by these specialists for imaging to exclude vascular TBT compression and were subsequently evaluated by a cardiovascular surgeon, who confirmed the diagnosis and determined the indications for repair.

Surgical treatment of a vascular ring is intended to completely relieve compression of the TBT and esophagus

Table 6. Chylothorax according to vascular ring type

Chylothorax	Double aortic arch, n (%)	Right aortic arch with left ligamentum, n (%)	Left aortic arch with right ligamentum, n (%)	Pulmonary artery sling, n (%)	p
Absent	9 (56.2)	29 (85.3)	3 (75.0)	4 (100.0)	0.089
Present	7 (43.8)	5 (14.7)	1 (25.0)	0 (0.0)	

and prevent future respiratory and interstitial lung disease. In some patients, manifestations of respiratory failure, such as dyspnea, pneumonia, and atelectasis, may not resolve completely because tracheobronchomalacia has already developed. This is generally attributable to delayed surgical treatment [4; 11; 14].

The widespread implementation of modern imaging techniques and a multidisciplinary approach to the diagnosis and treatment of vascular rings, previously considered a rare congenital condition, enabled us to accumulate experience with 58 patients over a relatively short period. Our findings indicate that, with an appropriate surgical strategy based on timely multimodal diagnosis and preoperative risk assessment, correction of vascular anomalies causing TBT and esophageal compression is effective and relatively safe.

Our review of the literature on surgical treatment of vascular rings identified no clear recommendations concerning the optimal age at repair. The reported mean age at surgery is 11–15 months [15; 16]. In our opinion, all types of vascular rings should be repaired before 1 year of age. Surgery at this age is technically easier, permits timely prevention of symptoms, and reduces the risk of tracheobronchomalacia. During the first year of life, symptoms can be readily confirmed, compression-induced changes in the TBT remain reversible, and the risk of perioperative complications, including chylothorax, is lower than in older children. Nevertheless, when severe symptoms and clinically significant TBT compression are confirmed by imaging, repair should be performed as soon as the anomaly is identified. Pulmonary artery sling should be corrected at the earliest possible time because all patients with this anatomical variant have severe baseline tracheal stenosis, which is frequently associated with the ring-sling complex [17].

The effect of age on complete symptom resolution requires further study. In our limited sample, no statistically significant association between patient age and symptom resolution could be demonstrated; therefore, this analysis was not included in the Results.

The indications for and timing of surgical treatment in asymptomatic patients with different forms of vascular rings remain controversial and require further investigation. Some authors advocate watchful waiting in asymptomatic patients. Others, including Oakley et al. and Sahu and Moe, have demonstrated substantial tracheal compression in the absence of symptoms [13; 18]. At our center, indications for surgery in asymptomatic patients are based on structural TBT abnormalities or clinically significant esophageal compression confirmed by MDCT, bronchoscopy, or contrast esophagography.

The advisability of aortopexy and its effects on long-term freedom from recurrent symptoms are of interest and require more thorough investigation. Some authors recommend adding aortopexy to division of the ligamentum arteriosum in all patients [19]. Others recommend diverticular resection with reimplantation of the left subclavian artery into the left common carotid artery in every patient with a diverticulum [20; 21]. Our study included patients who underwent neither left subclavian artery reimplantation nor diverticular resection. Aortopexy was used when the diverticulum was visually large, with a diameter at least 1.5 times that of the descending aorta, or visibly continued to compress the esophagus after division of the ligamentum arteriosum. Long-term follow-up involving symptom assessment and diagnostic surveillance will provide additional information on the effects of these different strategies.

Despite the absence of mortality and the high overall effectiveness of surgery, the high incidence of postoperative chylothorax (22.4%) warrants attention. According to published data, the incidence of chylothorax after intrathoracic surgery ranges from 0.59% to 6.00% [22]. Recent evidence, however, suggests a higher incidence specifically after vascular ring surgery. In a multicenter cohort study, chylothorax occurred after vascular ring division in 13.2% of patients, compared with 4.5% after all cardiac surgical procedures, including the Fontan procedure [23]. Our retrospective analysis identified no predictors of this complication, emphasizing the need to investigate the causes of chylothorax in this setting and improve surgical techniques and postoperative management protocols.

Conclusions

1. In the study groups, surgical correction of vascular rings in infants and young children resulted in complete symptom resolution during the early postoperative period in 83.7% of symptomatic patients.
2. A comprehensive multimodal diagnostic approach to vascular rings enables timely detection of tracheal, bronchial, and/or esophageal narrowing in asymptomatic patients and supports the formulation of indications for surgical treatment.
3. Aortopexy was performed most frequently in patients with a right aortic arch and left ligamentum arteriosum to relieve residual esophageal compression. The procedure was not associated with the incidence of chylothorax.
4. Chylothorax occurred during the early postoperative period in 22.4% of patients and was most frequent

in patients with a double aortic arch. It prolonged hospitalization, although no statistically significant between-group differences in incidence were found.

Study limitations

This was a retrospective, single-center study with a relatively small sample and imbalanced group sizes. We

evaluated the effects of surgical treatment based on symptom status during the early postoperative period and the occurrence of surgical complications. No comparison was made with a control group of children who did not undergo surgery. Long-term outcomes, including residual or recurrent symptoms, and standardized assessment of quality of life also require further investigation.

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