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# Thoracoscopic division of vascular rings in infants and children

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**Index words:**
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**Abstract**

**Objective:** Traditionally vascular rings in infants and children are treated through an open thoracotomy. Recently, thoracoscopic surgery has been used for these complex procedures. This study reports our early experience with thoracoscopic division of vascular rings and evaluates the efficacy and safety of this approach.

**Material and Methods:** Patients who underwent thoracoscopic division of vascular rings at King Khalid University Hospital, Riyadh, Saudi Arabia, from December 2004 to January 2006 are included. Their data were carefully analyzed looking at demographics, clinical presentation, diagnostic modality, type of the anomaly, operative details, complications, and outcome.

**Results:** A total of 9 patients underwent thoracoscopic division of vascular rings. Age at surgery ranged between 2 and 108 months (mean, 24 months). Weight varied between 5.3 and 32 kg (mean, 10.3 kg). All patients were symptomatic. Computed tomographic scan was diagnostic and accurately defined the type of anomaly in all the patients. Four patients had a right aortic arch with an aberrant left subclavian artery and left ductus/ligamentum arteriosum, 2 had double aortic arches, and 3 had a right aberrant subclavian artery. One patient developed right-sided pneumothorax on the contralateral site, and another one developed apnea 12 hours after surgery, requiring mechanical ventilation. There was no mortality. Operative time ranged between 50 and 145 minutes, the mean being 107 minutes. The average hospital stay was 4 days. Five patients had their preoperative symptoms completely resolved, and the rest are showing steady improvement. The average follow-up period is 6 months.

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**Conclusion:** Our early experience indicates that thoracoscopic division of vascular rings is safe and effective. Because it takes away the need for thoracotomy, it is likely that it can result in less postoperative pain and rapid convalescence. It also prevents the ill effects of thoracotomy and gives good cosmetic results.
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A vascular ring is an uncommon congenital anomaly that results from erroneous embryological development of the arterial component of the branchial arch system in which a complete or incomplete ring is formed around the trachea and esophagus. The symptoms associated with a vascular ring relate to the structure that is encircled by it, the trachea, the esophagus, or both.

Surgery when feasible is the treatment of choice. The accepted operative approach to a symptomatic vascular ring is the division of vascular ring through a posterolateral thoracotomy. The first successful division of a double aortic arch was done by Gross [1] in 1945. With the refinement of video-assisted thoracoscopic surgical techniques and miniaturization of instruments and video equipments, more procedures are being attempted thoracoscopically in children. The first video-assisted thoracoscopic division of a vascular ring was described by Burke and Chang [2] in 1993.

## 1. Materials and methods

We reviewed the records of patients who underwent thoracoscopic division of vascular rings at King Khalid University Hospital, Riyadh, Saudi Arabia, from December 2004 to January 2006. Their data were carefully analyzed looking at demographics, clinical presentation, diagnostic modality, type of anomaly, and operative details. The analysis of operative outcome included study of postoperative complications, duration of postoperative ventilation, length of hospitalization, and improvement in symptoms.

All the patients were symptomatic at the time of surgery. Presenting symptoms and signs were airway related (n = 1), dysphagia (n = 3), or dysphagia and airway related (n = 5); respiratory symptoms were aggravated by feeding in 5 patients. All the patients underwent a preoperative esophagogram that revealed posterior indentation of the esophageal wall in all patients. Chest computed tomographic (CT) scans with contrast and 3-dimensional (3D) volume rendering was obtained in all patients preoperatively. They were diagnostic, accurately defined the anatomy and the type of vascular anomalies, and greatly helped in planning of surgical strategy. Echocardiogram was done in 4 patients but was diagnostic only in 1.

## 2. Surgical technique

The procedure was performed with the patient in a lateral decubitus position with an added 15° to 20° in a slightly prone direction. Single-lung ventilation was achieved by selective intubation of the contralateral side in older children or by the use of bronchial blocker of the ipsilateral lung in small infants. Intrathoracic $CO_2$ pressure of 4 to 6 mm Hg was used to obtain more lung collapse in some cases. Central venous and arterial catheters were placed in all patients. Routine monitoring of bilateral upper-extremity transcutaneous oxygen saturation was obtained. In addition, serial arterial blood gas measurements, continuous end-tidal $CO_2$ monitoring, and blood pressure were periodically assessed. Continuous electrocardiogram was displayed. The standard 3-trocar technique was used. One 5-mm trocar was placed in the fifth intercostal space in midaxillary line to accommodate the 5-mm, 30° telescope. Two additional working ports were used; a 3-mm reusable trocar was placed in the third intercostal space at the anterior axillary line, and a 5-mm or 10-mm trocar was placed in the sixth intercostal space at the posterior axillary line to be used for dissection and clipping of the ring. All our procedures were carried out through a left thoracoscopic approach except for one with a double aortic arch (DAA) and a dominant left arch, in which we elected to use the right side.

The procedure was started by opening the mediastinal pleura and, usually, dividing the superior intercostal vein. The anatomy of the aortic arch and its relationship to subclavian artery was clearly displayed. Clarification of anatomy was greatly facilitated by carefully studying the 3D volume rendering CT scan preoperatively. The ring elements were dissected free off the underlying esophagus and surrounding structures.

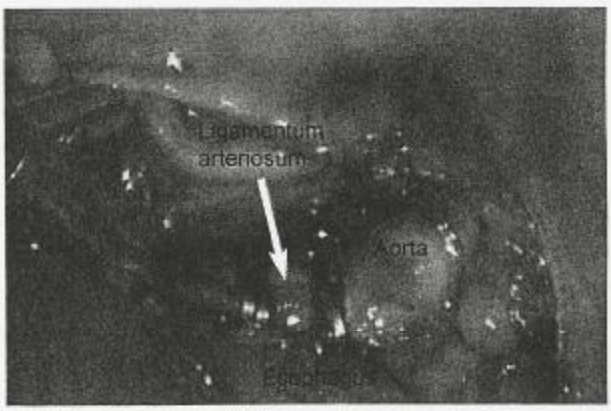


**Fig. 1** Thoracoscopic view of the left upper thoracic cavity showing vascular clips applied on the left ductus arteriosum in a patient with a right aortic arch and an aberrant left subclavian artery.

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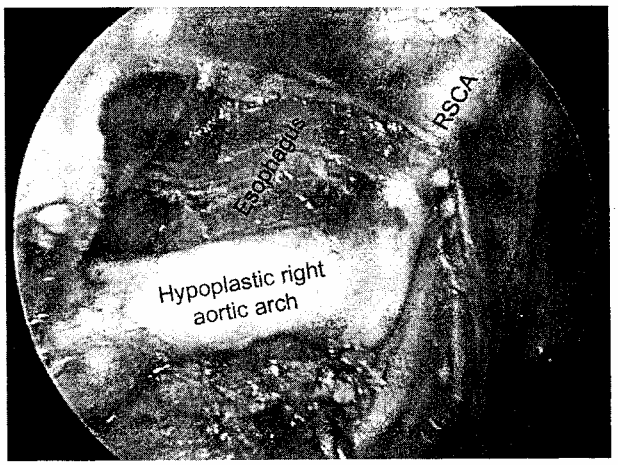


**Fig. 2** Thoracoscopic view of the right upper thoracic cavity showing a right hypoplastic patent arch before division in a patient with a DAA (dominant left arch).

In patients with a right aortic arch and an aberrant left subclavian artery, the ductus/ligamentum arteriosum was divided between vascular clips using a 5-mm or 10-mm clipper, depending on the size of the ductus (Fig. 1). In cases of a DAA, the hypoplastic patent arch was doubly clipped at each end distal to the subclavian artery and divided (Fig. 2). Fibrous bands along the esophagus were also divided. In patients with an aberrant right subclavian artery, the vessel was carefully dissected in its entirety from its origin distal to the left subclavian artery and was divided between vascular clips (Fig. 3). All of our patients had a chest tube placed under vision at the end of the procedure. In one case, however, we did not find this to be necessary.

## 3. Results

A total of 9 patients underwent thoracoscopic division of vascular rings during the period between December 2004 and January 2006. There were 4 boys and 5 girls. The mean age was 24 months (range, 2-108 months), and mean weight was 10.3 kg (range, 5.3-32 kg) (Table 1).

The types of vascular rings included DAA (n = 2), right aortic arch with aberrant left subclavian artery and left ligamentum/ductal arteriosum (n = 4), and aberrant right subclavian artery (n = 3). The total operative time ranged from 50 to 145 minutes (mean, 107 minutes). There was no conversion to open thoracotomy, and there were no intraoperative complications. The vascular anatomy was consistent with the preoperative studies in all patients. Chest tubes were placed in all patients except in one patient. They were removed between 24 and 48 hours postoperatively. Extubation was performed in 8 patients in the operating room and after 72 hours in 1 patient. Feeding started within 24 hours in 6 patients and after 24 hours in 3 patients. Two patients had postoperative complications, and 1 developed apnea after intravenous fentanyl, thereby needing reintubation. Another one developed contralateral pneumothorax, which is most likely because of barotrauma; he was managed by a chest tube insertion and did well. The mean hospital stay was 4 days (range, 2-12 days), and the length of follow-up ranged from 2 to 12 months (mean, 6 months). Five patients (3 with an aberrant right subclavian artery and 2 with a right aortic arch and an aberrant left subclavian artery) experienced immediate and complete resolution of their symptoms after surgery. The remaining 4 patients (2 with a DAA and 2 with a right aortic arch and an aberrant left subclavian artery) are showing progressive improvement in their symptoms over the follow-up period. Feeding difficulty and dysphagia were the first symptoms to disappear, whereas the respiratory problems are slower to abate. No patient was rehospitalized for recurrent chest infection or feeding difficulty.

## 4. Discussion

Vascular rings are uncommon anomalies in which the anomalous configuration of the arch and/or associated vessels surrounds the trachea and esophagus, forming a complete or incomplete ring around them. Symptoms and physical findings produced by vascular rings are primarily those of airway or esophageal compression. Children with narrow or tight rings have a significant degree of constriction of one or both of these structures and present very early in life. Most patients with vascular rings present with symptoms in infancy or very early in childhood. However, a small number of patients do not develop symptoms until later in life, whereas others remain entirely asymptomatic. Common symptoms include stridor; cyanosis; respiratory distress; apnea; and/or a characteristic high pitched, brassy cough. Additional findings include recurrent chest infections, history of asthma, and evidence of dysphagia or difficulty with feeding.

**Fig. 3** Thoracoscopic view of the left upper thoracic cavity showing an aberrant right subclavian artery after division between vascular clips.

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**Table 1** Characteristics of 9 patients who had undergone thoracoscopic division of a vascular ring

| No. | Age (mo) | Weight (kg) | Sex | Type of vascular ring |
|---|---|---|---|---|
| 1 | 5 | 6.8 | Male | RAA, aberrant LSCA, left ligamentum arteriosum |
| 2 | 30 | 11.5 | Male | RAA, aberrant LSCA, left ligamentum arteriosum |
| 3 | 36 | 7.9 | Female | RAA, aberrant LSCA, left ductus arteriosum |
| 4 | 16 | 9.3 | Female | RAA, aberrant LSCA, left ductus arteriosum |
| 5 | 4 | 65 | Male | Aberrant RSCA |
| 6 | 10 | 8.8 | Female | Aberrant RSCA |
| 7 | 2 | 5.3 | Female | Aberrant RSCA |
| 8 | 108 | 32 | Female | DAA with right arch dominant |
| 9 | 5 | 5.3 | Male | DAA with left arch dominant |

RAA indicates right aortic arch; LSCA, left subclavian artery; RSCA, right subclavian artery.

Some have the impression that children with dysphagia lusoria because of an aberrant right subclavian artery may outgrow their symptoms and are best managed expectantly. In our experience and that of others, this is not always the case [3,4]. All our patients with this anomaly were indeed symptomatic, and the link between this abnormality and the patients' symptoms was clearly established preoperatively. The other point of equal interest is the significant improvement in the symptoms after surgery.

When entertaining the possibility of a vascular ring, the question of the best means of diagnosis emerges. Our data and those of others [3,4] have shown that an esophagogram remains an excellent means for demonstrating the presence of a vascular ring. Perhaps the most attractive imaging modality available is the use of CT or magnetic resonance imaging [5-7]. Computed tomographic scan with contrast 3D volume rendering is a safe, fast, and noninvasive method for diagnosis. It provides most of the necessary information about vascular rings that is not provided by other techniques. The 3D reconstructed images obtained with 3D CT allow the surgeon to visualize the vessels responsible for the compression and analyze both its size and the arteries that arise from the aortic arch, thus helping to choose the best operative approach and plan. In all of the patients, 3D CT scan with volume rendering accurately diagnosed and defined the type of vascular ring. It helped us to select the appropriate thoracic cavity for entry to best deal with a hypoplastic arch in patients with a DAA. This was evident in one of our cases, where we approached a patient with a DAA through the right side because 3D CT scan revealed that the hypoplastic arch was in the right side.

The accepted operative approach to a symptomatic vascular ring has been an open surgical procedure through a posterolateral thoracotomy. With the refinement of video-assisted thoracoscopic surgical (VATS) techniques and miniaturization of instruments and video equipments, VATS is a viable alternative to open thoracotomy for management of the symptomatic vascular ring in children. In 1993, Burke and Chang [2] reported the first case of thoracoscopic division of vascular ring. Subsequently, in 1995, Burke et al [8] reported their series of 8 patients with vascular rings that were divided by using VATS techniques. There were 3 conversions to a regular thoracotomy in that series. In 2005, Koontz et al [9] published a series of 13 patients with vascular rings that were thoracoscopically divided. Most of his patients had a right aortic arch with an aberrant left subclavian artery and left ligamentum arteriosum, which is very similar to our series. Mihaljevic et al [10] have reported the first use of a telerobotic surgical system (Da Vinci system, Intuitive Surgical, Sunnyvale, CA) for the division of vascular rings in 2 children.

Important steps for a successful thoracoscopic division of a vascular ring should include careful evaluation of a preoperative imaging study to provide clear surgical approach and preoperative strategy, proper port placement, adequate lung collapse, careful dissection to display the anatomy of the aortic arch and great vessels, double clipping of the ring vessels at each end before division, dividing the constricting fibrous bands around the esophagus, and lastly, ensuring rapid access to the thoracic cavity in the event of bleeding. The other option for dealing with these vessels is double ligation at each end before division; although we have not used this option, it sounds feasible. In children, the aberrant right subclavian artery may simply be clipped or ligated and divided without sequalae. A subclavian steal syndrome may result after simple division in adults, so reimplantation to the aorta may be necessary [11].

In our opinion, surgeons who are experienced in handling vascular rings by the conventional open technique will be able to do it safely thoracoscopically, provided they acquire adequate experience of the VATS technique. Although the follow-up of our patients is rather short (2-12 months), more than half of them have their symptoms completely resolved, and the rest are showing progressive improvement. This is very encouraging and indeed is similar to what one would expect from the open technique. Bonnard et al [4] reported complete improvement in 68% of cases, partial improvement in 17%, and no improvement in 15% of cases that were treated by open thoracotomy.

Our early experience indicates that thoracoscopic division of vascular rings is safe and effective. Because it takes away the need for thoracotomy, it is likely that it can result in less pain and rapid convalescence. It also prevents the ill effects of thoracotomy and gives good cosmetic results.

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