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Video assisted thoracoscopic surgery cases with right-sided aortic arch aneurysm and complete vascular ring: Case report

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ABSTRACT

INTRODUCTION: Right-sided aortic arch with aberrant left subclavian artery and ligamentum arteriosum, after double aortic arch, is the second most common complete vascular ring. It was traditionally treated by open surgical thoracotomy and recently video assisted thoracoscopic surgery (VATS) has been used in some cases.

PRESENTATION OF CASE: We describe the cases of two infants who presented with gastroesophageal reflux, dyspnea, dysphagia secondary to aneurysmal dilatation of the retroesophageal arch confirmed by imaging data. VATS procedure was performed through a left thoracoscopic approach. Ligamentum arteriosus compressed esophagus was clipped, sectioned and then released the esophagus in one case; also, In the second case, we clipped and sectioned aorta, distal to the origin of aberrant left subclavian artery.

DISCUSSION: CT angiography and MRI are known to be the most effective available imaging methods for vascular ring detection. Also, there are several surgical approaches to vascular rings such as, thoracotomy and thoracoscopy. There is a large body of evidence confirming the safety, efficacy and convenience of VATS as a therapeutic option for congenital heart disease including right-sided aortic arch and aberrant left subclavian artery.

CONCLUSION: VATS is a less invasive and safe strategy for management of right-sided aortic arch with aberrant left subclavian artery and ligamentum arteriosum.

1. Introduction

One of the rarest congenital cardiac defects is the complete vascular ring.[1] Vascular ring is caused by failure of involution or migration of particular sections of the aortic arches, specially, abnormal involution of the left fourth arch which leads to a right sided aortic arch with retroesophageal left subclavian artery and ligamentum. This variation is one of the most prevalent complete vascular rings after double aortic arch.[1] However, true aneurysmal dilatation of the aortic arch is rare and a Kommerell diverticulum may be correlated with the origin of the left subclavian artery.[2]

Thoracoscopic division of a vascular ring was first introduced by Burke in 1993 and has become progressively popular in the treatment of vascular rings due to its safety and low complications such as decreased surgical trauma, reduced intensive care unit (ICU) and hospital stay, an enhanced recovery, and an improved cosmetic result.[3,4] Recently, the use of video-assisted thoracoscopic surgery) VATS) has been increasingly popular in the treatment of congenital heart defects. However, due to the proficiency-dependence and increasing cost, this is rarely used in developing countries.[5] We report two cases of right-sided aortic arch with aneurysmal dilatation of its retroesophageal segment treated with VATS.

2. Case reports

2.1. Case 1

A 7-month-old female infant with vomiting after feeding, stridor, high pitched and brassy cough, noisy respiration, emesis and respiratory distress at birth was referred to our institution. Early evaluations presented gastrointestinal reflux disease (GERD). Also, further diagnostic tests such as barium swallow test (Fig. 1) and computed tomography (CT) Angiography (Fig. 2) were done for congenital vascular abnormalities. These two tests demonstrated that there were round compressive lesions posterior to the esophagus, right sided aortic arch with aberrant left subclavian artery (LSA), Kommerell's diverticulum and ligamentum arteriosum.

2.2. Case 2

A 15-month-old female infant born with the history of respiratory distress right after birth who needed to be under lung

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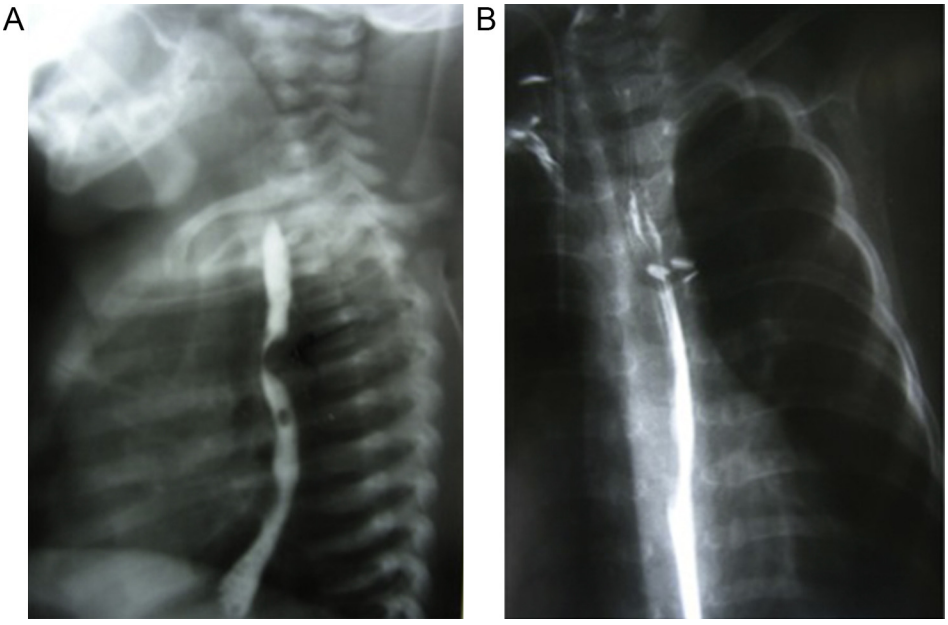

Fig. 1. (A) Round compressive lesion, narrowing the esophagus – before surgery (Case 1). (B) Normal diameter without narrowing of the esophagus – after surgery (Case 1).

ventilation for 2 weeks was referred to our institution. Every attempt at extubation resulted in an instant respiratory distress. A diagnosis of the aortic arch and vascular ring was made with cardiac dynamic magnetic resounance imaging (MRI) with gadolinium. Imaging data showed right-sided aortic arch with separate origin of all supra aortic branches, left superior vena cava (LSVC) which drained into dilated coronary sinus, right superior venacava without innominate bridge to LSVC and normal left and right ventricular sizes. Right-sided aortic arch with aberrant left subclavian artery in addition to the left aortic arch formed a vascular ring.

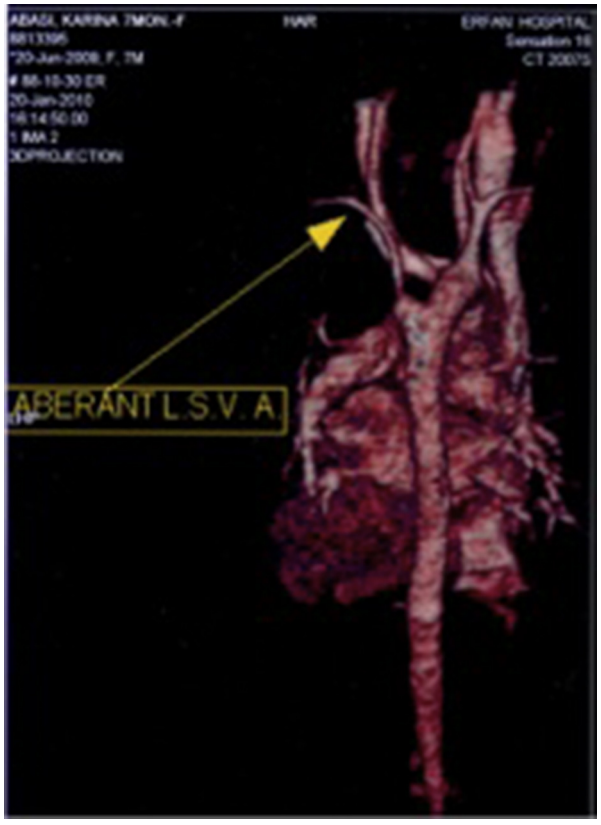

Fig. 2. CT angiography before surgery (Case 1).

The operative approach for both cases was planned with VATS procedure. Prior to surgery, central venous and arterial catheters were placed in all patients. Procedure was performed through a left thoracoscopic approach. A standard 3-trocar technique was used. One 5-mm trocar was placed in the fifth intercostal space in mid-axillary line to accommodate the 5-mm, 0° telescope. The ring elements were dissected and freed from the underlying esophagus and surrounding tissues. Then, the ligamentum arteriosum in case 1 and distal to the abberant left subclavian artery in case 2 (Fig. 3) were divided between vascular clips using a 5 mm and 10 mm clips, respectively. Fibrous bands along the esophagus were also divided. Patients had a chest tube placed under vision at the end of the operation, the pleural catheter was removed under positive pressure and previously placed suture was tied. After performing VATS procedure, the barium swallow test results, suggested normal diameter and peristalsis without any narrowing of esophagus (Fig. 1). In addition, all the symptoms had disappeared by the time of discharge from the hospital.

3. Discussion

After double aortic arch, right-sided aortic arch with left ligamentum arteriosum and aberrant LSA are the most common congenital cardiac defects which are usually characterized by esophageal or airway narrowing in the early years of life.[2] These symptoms may vary from GERD and apnea to difficulty in swallowing, respiratory distress and high-pitched cough in some situations.

CT angiography and MRI are the most effective available imaging methods. The chest radiography illustrated an abnormal aortic arch pattern or narrowing of the tracheal shadow, suggesting the presence of vascular ring.[6] CT scan with contrast 3D volume is a rapid, safe and somewhat noninvasive imaging which is used for diagnosis and preoperative evaluation of vascular ring that enables surgeons to visualize the vessels which take part in the constriction and therefore it helps to choose the best plan for VATS.[7] Upper gastrointestinal series is a supplemental diagnostic technique that helps to prove a vascular ring by showing posterior indentation of

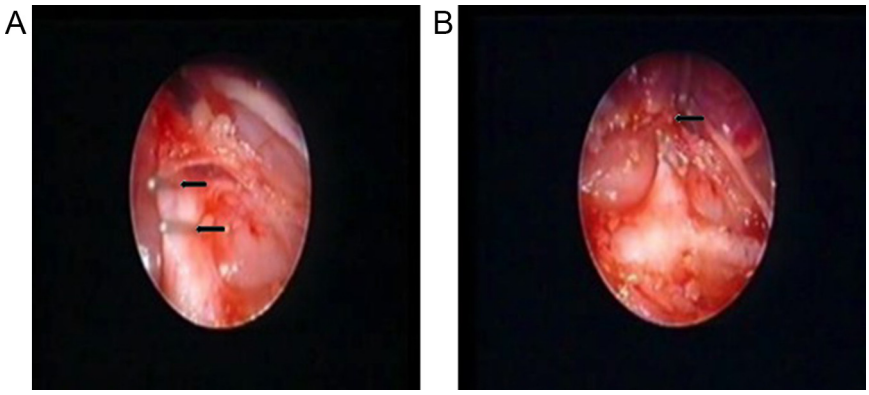

Fig. 3. (A) Right-sided aortic arch with abberant left subclavian artery – clipped (Case 2). (B) Right-sided aortic arch with abberant left subclavian artery – divided (Case 2).

the esophagus from blood vessels passing behind it.[6] In our cases, complete vascular ring was diagnosed by chest radiography, cardiac MRI, 3D CT angiography and esophagography.

There are several surgical approaches to vascular rings such as, thoracotomy and thoracoscopy. In the left thoracotomy procedure, access to the ascending aorta and its branches is an important problem.[8]

There is a large body of evidence suggesting effectiveness, safety and convenience of VATS and its excellent outcomes; especially in patients with right aortic arch and aberrant left subclavian artery.[5,9–11] However, some studies have concluded that there is no significant difference in patients' outcome and the duration of operation between VATS and the traditional thoracotomy approach.[4,7] Some other studies have revealed greater operative time in thoracotomy approach.[9,10]

Kogon et al. observed that there are no significant differences in operative time among patients who had undergone thoracotomy and those who had undergone VATS for vascular ring. However, patients with the thoracoscopic approach experienced short length of ICU stay and hospital stay in comparison with those with thoracotomy approach.[4]

Some issues about the possible variation of performing VATS on the vascular ring are still controversial. These include preoperative imaging, type of vascular ring, the use of CO_2 insufflation, and the importance of performing a concomitant diverticulopexy. We believe that resection of Kommerell's diverticulum is rarely necessary, although some centers routinely do this, believing that greater relief of potential reobstruction or aneurysm formation is possible.

4. Conclusion

VATS is a less invasive and safe strategy for management of right-sided aortic arch with aberrant left subclavian artery and ligamentum arteriosum.

Conflict of interest

The authors declare that they have no conflict of interest.

Ethical approval

The study was conducted in accordance with the principles of Declaration of Helsinki 1996 version and Good Practice standards. All subjects signed informed-consent forms.

Authors' contributions

Mohammad Hassan Nezafati: Head of research team, writing. Pouya Nezafati: data collection, writing.

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