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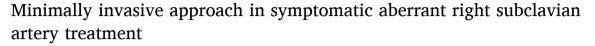
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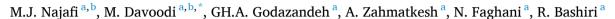
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Case report





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ABSTRACT

Introduction and importance: Anomalous right subclavian artery (ARSA) represents an uncommon anatomical deviation concerning the genesis of the right subclavian artery. As the predominant embryological irregularity of the aortic arch, it is clinically recognized as arteria lusoria (AL).

Case presentation: This study, describe the instance of a 22-year-old female exhibiting a non-aneurysmal, symptomatic anomalous right subclavian artery (ARSA) coursing posteriorly to the esophagus, as evidenced by thoracic computed tomography (CT) imaging.

Clinical discussion: As an attractive option, a minimally invasive surgical method was used to treat the patient, and the anomalous vessel was closed from the closest location to its origin in the aortic arch during a short time thoracoscopic surgery.

Discussion, conclusion: Compared to the common surgical methods to treat this anomaly, the complications and morbidity resulting from this method are much less and the length of stay in the hospital is shorter and the results are acceptable.

1. Introduction

Anomalous right subclavian artery (ARSA) constitutes an infrequent anatomical deviation regarding the genesis of the right subclavian artery. As the most prevalent embryological irregularity of the aortic arch, it is clinically referred to as arteria lusoria (AL) [1]. Typically, the right subclavian artery arises from the brachiocephalic artery; however, in 0.4–1.8 % of the general populace, this vessel originates as the final branch of the aortic arch, beyond the left subclavian artery [2,3]. Proceeding towards the right arm, it traverses the body's midline, positioned between the esophagus and vertebral column in 80 % of cases, between the trachea and esophagus in 15 % of cases, or even anterior to the trachea in 5 % of cases [4]. Should the artery exert pressure on the esophagus, it may result in a condition termed dysphagia lusoria [2–4]. Frequently, the arteria lusoria arises from an aortic arch diverticulum at the proximal descending aorta, first described by Kommerell [5].

This anatomical irregularity may be correlated with several clinical symptoms, including dyspnea, stridor, dysphagia (referred to as dysphagia lusoria), chest discomfort, or fever [6–8]. but majority of cases with ARSA are asymptomatic.

In diagnostic workup, several options are available, including upper gastrointestinal tract examination with barium-contrast series, which can rule out esophageal abnormalities, but computed tomography (CT) and magnetic resonance imaging (MRI) are considered the main diagnostic modalities, which provide the possibility of a detailed examination of ARSA and surrounding structures with further information regarding any aneurysmal degeneration [9].

Surgery is indicated when patients become symptomatic. Generally, the open surgery with right subclavian artery bypass performed, but today less invasive approach is popular in medical managements and also in this filed, so this article present less invasive approach to treat symptomatic aberrant right subclavian artery.

This case present based on the SCARE guideline [10].

2. Presentation of case

A 22-year-old right-handed Caucasian female, standing 145 cm tall and weighing 40 kg, reported experiencing challenges in consuming solid food over the past two years. This issue progressively worsened, and during the previous year, she also developed difficulty in

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swallowing liquids. Additionally, she experienced a weight loss of approximately 6 to 8 kg within the last six months. In her past medical history, she had congenital cataract, left kidney surgery in 5 years old due to congenital kidney cyst, cholecystectomy in 9 years old, brain haemangioma in third ventricle and Lung cyst.

Upper gastrointestinal endoscopy was negative for an esophageal mass and barium-contrast examination of the upper gastrointestinal tract shows well define defect in upper esophagus due to compression/compressing effect on upper esophagus (Fig. 1a and b), so manometry was performed that reported a high-pressure band at proximal part of esophagus at 28 cm with approximal 80 mmHg pressure with bolus transit.

A contrast-enhanced CT scan showed ARSA originating from the left aortic arch without aneurysmal dilatation and bypassing the thoracic esophagus posteriorly (Fig. 2a and b).

A minimally invasive surgical approach was performed for patient due to persistent symptoms and complaints. Under general anesthesia and double-lumen endotracheal tube, the patient was placed in the left lateral decubitus position. Through a right triportal thoracoscopic approach, the ports were placed in the fifth and eighth intercostal space. The mediastinal part of parietal pleura close to distal portion of the aortic arch was opened allowing an adequate exposure of the ARSA: the anomalous vessel, dissected free from the surrounding tissue, was encircled by a vessel loop and divided at its origin at the aortic arch with an endovascular stapling device (Fig. 3a and b).

Throughout the course of the surgical procedure, the patient exhibited stable vital signs without any incidences of hemorrhage. Upon completion of the surgery, positive pulses were noted in the right radial artery, and the arterial line reflected normal blood pressure levels in the right upper extremity. Additionally, normal oxygen saturation levels were confirmed, negating the need for a right subclavian artery bypass. The operation, which concluded after approximately one hour, included the placement of a chest tube for pleural drainage.

Following a two-day post-surgical stay, the patient was discharged with a Heimlich valve, necessitated by a significant air leak attributable to lung cysts. In the ensuing postoperative period, the patient demonstrated normal swallowing function. A contrast-enhanced CT scan

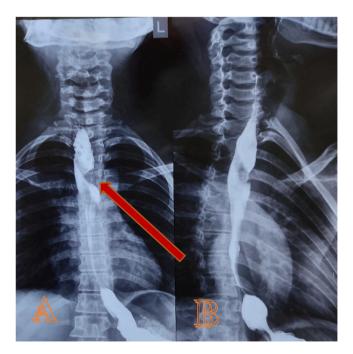


Fig. 1. a and b Barium-contrast examination of the upper gastrointestinal tract: (A) a well define defect in upper esophagus due to compression/compressing effect on upper esophagus (B) lateral view.

conducted two weeks post-surgery revealed no signs of aneurysmal degeneration in the residual aberrant right subclavian artery (ARSA) stump (Fig. 4).

3. Discussion

The initial account of this variation was documented by Hunauld in 1735 [8]. Nevertheless, the clinical manifestation of "dysphagia lusoria" was first delineated by Bayford in 1787, following the examination of a female presenting a lengthy history of dysphagia, who was discovered to possess an anomalous right subclavian artery upon autopsy [11]. Consequently, this condition is also referred to as Bayford-Autenrieth dysphagia. Almost in most cases the anomaly is asymptomatic, but sometimes it's associated with various signs or symptoms, that dysphagia is the most frequent complaint [12]. As per existing literature, arteria lusoria compression symptoms are observed in merely 7-10 % of adult patients exhibiting the anomaly. Thus, the anomaly remains clinically asymptomatic in 90-93 % of instances [13]. Given that the aberrant artery may traverse the space between the esophagus and vertebral column or the gap between the esophagus and trachea along the midline, it can potentially lead to dysphagia. Three distinct factors are hypothesized to contribute to the manifestation of symptoms in ARSA: elongation of the aberrant artery around the esophagus, increased sclerotic and reduced plastic nature, particularly in older individuals, or the presence of an aneurysm [7]. The prevalence of an anomalous right subclavian artery is comparatively greater in individuals with conditions such as Down's, DiGeorge, and Edwards' syndromes, as opposed to the general population [14]. Nakajima et al. [15] reported an incidence of 6 % for aberrant subclavian arteries in patients with tetralogy of Fallot, and 16 % in those presenting with a combination of pulmonary atresia or major aorticopulmonary collateral arteries. Furthermore, de Luca et al. [16] documented that, out of 12 diagnosed patients with ARSA, 6 also exhibited Down's syndrome, ventricular septal defects, and tetralogy of Fallot.

Individuals exhibiting ARSA along with symptoms associated with esophageal or tracheal compression, or those presenting with aneurysmal dilation, necessitate surgical intervention. Approaches such as median sternotomy, left thoracotomy, or bilateral carotid-subclavian bypasses followed by thoracic aortic endograft are recommended for cases involving aneurysmal disease. For patients with non-aneurysmal, symptomatic ARSA, the therapeutic objective entails the occlusion of the arteria lusoria origin and, if required, the revascularization of the right subclavian artery [17].

Certain literature sources have documented open ligation and transposition of the ARSA to the right common carotid artery via a right supraclavicular incision [18]. In order to circumvent potential aneurysmal formation stemming from a residual elongated stump of the anomalous vessel at its origin, a mediastinoscopy-assisted ligation of the ARSA, conducted through a right supraclavicular approach, has been recently suggested to enhance visualization of the operative area [19]. Nevertheless, this procedure can prove to be exceptionally challenging, given that the arteria lusoria origin is situated within a confined and hard-to-access anatomical region [18]. The potential for intraoperative hemorrhaging and the likelihood of leaving extended residual vascular stumps could be mitigated via hybrid endovascular techniques, such as the method proposed by Shennib et al. This approach involves conducting an end-to-side anastomosis between the severed distal ARSA and the right common carotid artery, prior to positioning an occlusion device 2 cm beyond the origin of the anomalous vessel [20,21].

4. Conclusion

Management approaches such as endovascular interventions are predominantly employed for elderly patients with comorbidities; however, there is a paucity of data regarding the long-term safety of endovascular occlusive devices in the literature. The minimally invasive

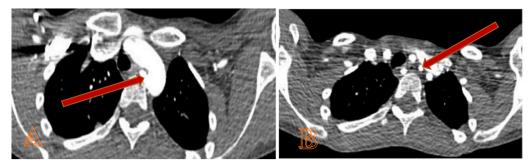


Fig. 2. a and b A contrast-enhanced CT scan performed for surgery planning and shows: (A) a non-aneurysmal arteria lusoria (red arrow) and (B) compressed and narrowed esophagus (red arrow).

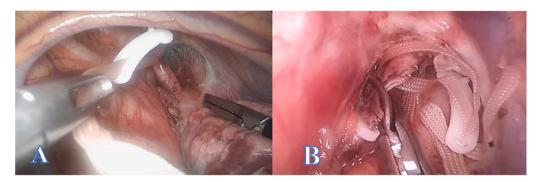


Fig. 3. a and b Intraoperative view of video-assisted thoracoscopic surgery. (A) Isolation of the arteria lusoria close to the aortic arch with vessel loop. (B) Mechanical stapling device for closure of the arteria lusoria. ARSA: aberrant right subclavian artery; RCCA: right common carotid artery.

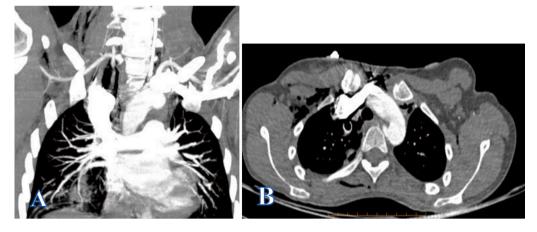


Fig. 4. a and b Preoperative contrast-enhanced CT scan shows: a non-aneurysmal arteria lusoria stump after surgery.

surgical technique executed in this case study facilitates a secure closure of the ARSA at its origin on the aortic arch through a right thoracoscopic procedure, eliminating the likelihood of leaving an extended residual vascular stump.

Patient (parent's) consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Ethical approval

This study was presented in the research ethics committee of Imam

Khomeini Hospital, Mazandaran University of Medical Sciences, Sari, Iran and waived due to the low risk assessment.

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There is nothing to declare.

Author contribution

Dr Mohammad Javad Najafi; writing the paper, surgery team and interpretation

 $\overline{\text{Dr}}$ Mahdi Davoodi; study concept and design, surgery head team and interpretation

Dr Gholam Ali Godazandeh; surgery team and interpretation Dr Ahmad Zahmatkesh; surgery team Dr Nafiseh Faghani; anesthesia head team Dr Reza Bashiri; surgery team

Guarantor

Dr Mahdi Davoodi; Vascular Surgeon, Mazandaran University of Medical Sciences

Dr Mohammad Javad Najafi; General Surgery Resident, Mazandaran University of Medical Sciences

Research registration number

N/A.

Conflict of interest statement

There is no conflicts of interest.

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