Three Cases of a Rare Association: Double Aortic Arch

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Abstract

Background
Vascular ring is less than 1% of congenital heart disease. Double aortic arch (DAA) is the most common form of it. Its detecting is important because of the effects of pressure on the esophagus and trachea.

Case Report
In this study, three children suffering from double aortic arch with symptoms of dysphagia and recurrent aspiration, which in two cases had led to cardiac arrest, were introduced. In all of these studies, computed tomography angiography and chest X-ray were used to diagnose the compression effect of double aortic arch on the esophagus and the surgery provided for treatment of the children. After the surgery, all three patients were monitored in the intensive care unit (ICU) for five days without showing any symptoms.

Conclusion
Totally, we found that three computed tomography angiography, as a non invasive, high quality method, would be suitable for the diagnosis of this abnormality.

Key Words: Aspiration; Children; Double aortic arch; Dysphagia.


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1-INTRODUCTION

Vascular ring, which comprises less than 1% of congenital heart disease, was first introduced by Gross in 1945 (1). As we know, double aortic arch is the most common form of vascular ring, which may cover the trachea and esophagus and thus cause the compression of these structures. If the compression is severe, symptoms such as shortness of breath, recurrent pneumonia, stridor, dysphagia and nutritional problems may be observed at birth. In this study, three children suffering from double aortic arch who had a history of foreign body in the trachea as well as dysphagia were reported.

2-CASE PRESENTATION

2-1. Case 1:

This case was a girl (1.5 years old), who has come with history of recurrent foreign body aspiration in the trachea (3 times) and dysphagia. Her body weight was 8.3kg. Her mother explained her poor feeding due to respiratory distress on feeding. She had history of cardiac arrest after respiratory suffering 2 weeks ago. After routine examinations chest x ray has done for her which was clear. The pediatric surgeon has suspected to vascular ring and requested angiography confirmed.

2-2. Case 2:

This case was a girl (2 years old), who has come with cyanosis and signs of acute distress to children hospital. Lung examination wasn’t showing any positive findings including foreign body aspiration in the trachea. Pediatric surgeon had done rigid bronchoscopy in operation room. Trachea and main bronchioles were intact. She was observed for 2 days and requested angiography confirmed vascular ring.

2-3. Case 3:

This case was a boy (2.5 years old) who had come with emergency service to children emergency department after successful resuscitation following cyanosis and cardiac arrest. His mother reported acute choking after eating bit of fruits. No foreign body aspiration found in rigid bronchoscopy in operation room and angiography showed double aortic arch. As can be seen in (Figure 1), in all three cases the anterior arch was smaller than the posterior arch. The surgeon then decided to perform a surgery; during the surgery, the surgeon both cut the ligature and attempted to block it since there were open ducts arteriosus in all three cases. After the surgery, all three patients were monitored in the intensive care unit (ICU) for five days without showing any symptoms.

Fig.1: Double aortic arch. The posterior arch is shorter that the anterior arch
3-DISCUSSION

As we know, one of the less common causes of persistent wheezing, recurrent pneumonia and dysphagia in children is vascular rings. Congenital abnormalities of the aortic arch and its main branches cause the formation of vascular rings around the trachea and esophagus and exert different levels of compression effect (2). Double aortic arch is the most common congenital vascular ring. The ascending aorta is divided into two branches before reaching trachea with the artery branches surrounding the esophagus and the trachea thoroughly (3). Although this vascular disorder is asymptomatic in some cases and is usually diagnosed incidentally in CT scan or adult angiography, or causes dysphagia, its clinical symptoms typically include stridor, coughing, wheezing and recurrent respiratory infections in infants and children (4). What was reported in the case presented in this study was that these children had dysphagia and recurrent aspiration, without displaying any symptoms of pneumonia.

Although most patients with double aortic arch show symptoms of tracheoesophageal obstruction, the results of the study conducted in 1968 indicated that among 11 cases of double aortic arch, 9 cases had no signs or symptoms (5). Consequently, the signs and symptoms of obstruction of the trachea and esophagus may be overlooked or interpreted as the clinical features of a severe internal injury. In recent years, however, DAA has been frequently cited in many studies and its management has been well-documented (6, 7).

4-CONCLUSION

As it was already mentioned in Sariaydin’s study, DAA (4), as a congenital abnormality may cause symptoms produced by compression effect on esophagus and trachea. In these patients, the surgery may be performed; otherwise, the follow-up treatment is strongly recommended. Three-dimensional CT scan and three CT angiography, as a noninvasive, high quality method, would be suitable for the diagnosis of this abnormality.

5-CONFLICT OF INTERESTS: None.

6-ACKNOWLEDGMENT

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7-REFERENCES