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Treatment of Dysphagia Lusoria with Supraclavicular Incision in Children: Aberrant Right Subclavian Artery

Çocuklarda Supraklaviküler İnsizyon İle Disfaji Lusoria Tedavisi: Sağ Aberran Subclavia

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ABSTRACT

Aberrant right subclavian artery (ARSA) is a rare (0.5-1.8% of the population), usually asymptomatic congenital anomaly. We report the three female patients who were successfully treated by surgery of supraclavicular incision. The median age at the operation time was 15 years old (range: 10-18 years). There were no peroperative and postoperative complication. Ulnar and radial tensions were bilaterally normal rate after surgery. There were no difficulties after the postoperative feeding. It may be suggested that end to side anastomosis of ARSA to carotid artery via supraclavicular incision is a feasible and minimal invasive method for children with dysphagia lusoria.

Key Words: Aberrant right subclavian artery, Dysphagia lusoria, Supraclavicular incision

ÖZ

Sağ aberrant subclavian arter (ARSA), nadir görülen ve genellikle asemptomatik seyreden konjenital bir anomalidir (populasyonun %0.5-1.8'i). Bu çalışmada supraklavikular insizyon ile başarılı bir şekilde tedavi edilen üç kız hastanın sunulması amaçlandı. Ameliyat sırasında ortanca yaş 15'di (aralık: 10-18 yaş). Ameliyat sırasında ve sonrasında komplikasyon izlenmedi. Bilateral ulnar ve radyal pulsasyonları ameliyat sonrası normaldi. Ameliyat sonrasında beslenme ile ilgili problem gözlenmedi. Supraklaviküler insizyon ile ARSA' nın uç yan anastomozu, disfaji lusoriası olan çocuklar için uygulanabilir ve minimal invaziv bir yöntem olarak düşünülmelidir.

Anahtar Sözcükler: Aberran sağ subklavian arter, Disfaji lusoria, Supraklavikular insizyon

INTRODUCTION

Among the dysphagia causes in children, aberrant right subclavian artery (ARSA) is a rare condition and it occurs to be 0.5-1.8 % of the population and may be asymptomatic (1). When it is symptomatic, children usually suffer from upper respiratory symptoms like stridor while adults suffer from dysphagia and chest pain (2). Dysphagia is presented when the ARSA compresses esophagus (1, 3, 4). Symptomatic patients require surgical treatment. Surgical approaches are median sternotomy, thoracotomy and supraclavicular incision (5). The best method has not been decided yet (3). The aim of this study to present three children with this rare reason for dysphagia that may easily be skipped.

CASE REPORT

Charts of three patients who underwent surgery because of dysphagia caused by ARSA between 2012 and 2015 were evaluated retrospectively. The underlying pathologies, preoperative preparations, surgical procedure, complications and dysphagia score were analyzed (6). Barium-contrast examination, angio-computed tomography and esophagogastroduedonoscopy were performed preoperatively for differential diagnosis and confirmed the presence of ARSA. Children were given water-soluble contrast orally (Urografin®). Extrinsic compression due to ARSA was demonstrated in all children (see Figure 1). All children with ARSA suspicion were performed Angio-Computed Tomography to evaluate vascular

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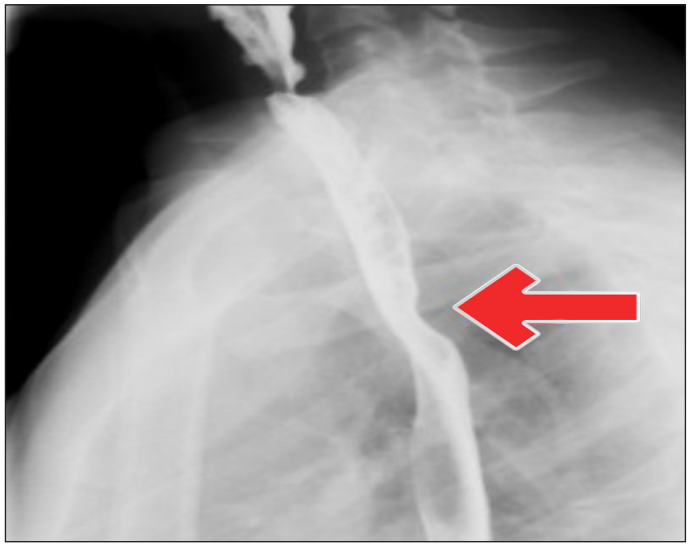

Figure 1. Contrast examination shows the extrinsic compression of ARSA.

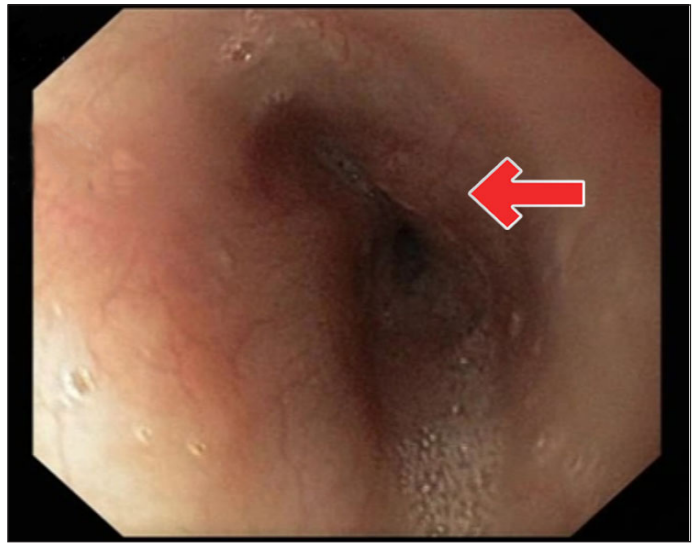

Figure 2. Pulcatile mass at posterior esophagus.

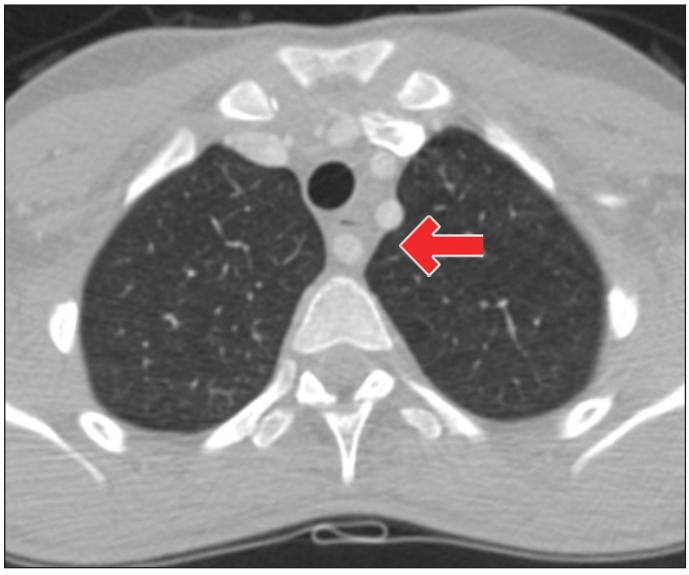

Figure 3. Angio-computed tomography showing the ARSA.

structures in detail and to be aware of any other vascular anomalies (Figure 2). Under general anesthesia, esophago-gastro-duedonoscopy was performed. Posterior portion of the esophagus was pulsatile because of extrinsic compression if there was ARSA that caused the dysphagia (Figure 3).

Surgical Technique

All three patients underwent surgery via right supraclavicular incision. A pillow was placed under neck to get extension in supine position. After incision to right sternocleidomastoid muscle and revealing right jugular vein, scalene muscles were cutting. The right subclavian artery was dissected to aortic arch. A vascular clamp was applied to the right subclavian artery at aortic arch junction and incision was made (Figure 4). Aortic side was repaired with 4/0 prolene and end to side anastomosis was performed to right carotid artery with 4/0 prolene.

Of three children who were included the study; all of them were female. The median age at the operation time was 15 years (range: 10-18 years). All children were presented with dysphagia. There were no peroperative and postoperative complication. Blood pressures which were measured from bilateral ulnar and radial arteries were normal after surgery. The mean hospital stay time was 9 days (7 days-12 days). Mean follow-up time was 4 years (2 years-5 years).There was no difficulties after the postoperative feeding.

DISCUSSION

ARSA is a rare cause of dysphagia (5). The most important step of a successful treatment is to think of ARSA in differential diagnosis. Afterwards ARSA can be managed in a feasible and minimally invasive fashion. Even though there are different surgical approaches and no consensus, end to side anastomosis to carotid artery via supraclavicular incision seems an appropriate method (3, 5).

ARSA is a rare (0.5-1.8% of the population), usually asymptomatic congenital anomaly (1). The embryologic abnormality which is responsible is the involution of fourth vascular arch (7). Even though this anomaly is usually asymptomatic, it may cause respiratory symptoms, dysphagia or chest pain due to the compression of trachea or esophagus (8). Even though respiratory symptoms are more common in children, they may be misdiagnosed in early childhood and the children may present with dysphagia in late childhood period as the 10 years old patient in this study. The dysphagia caused by ARSA first described by David Bayford in 1794 and may also referred to as dysphagia lusoria (8-10).

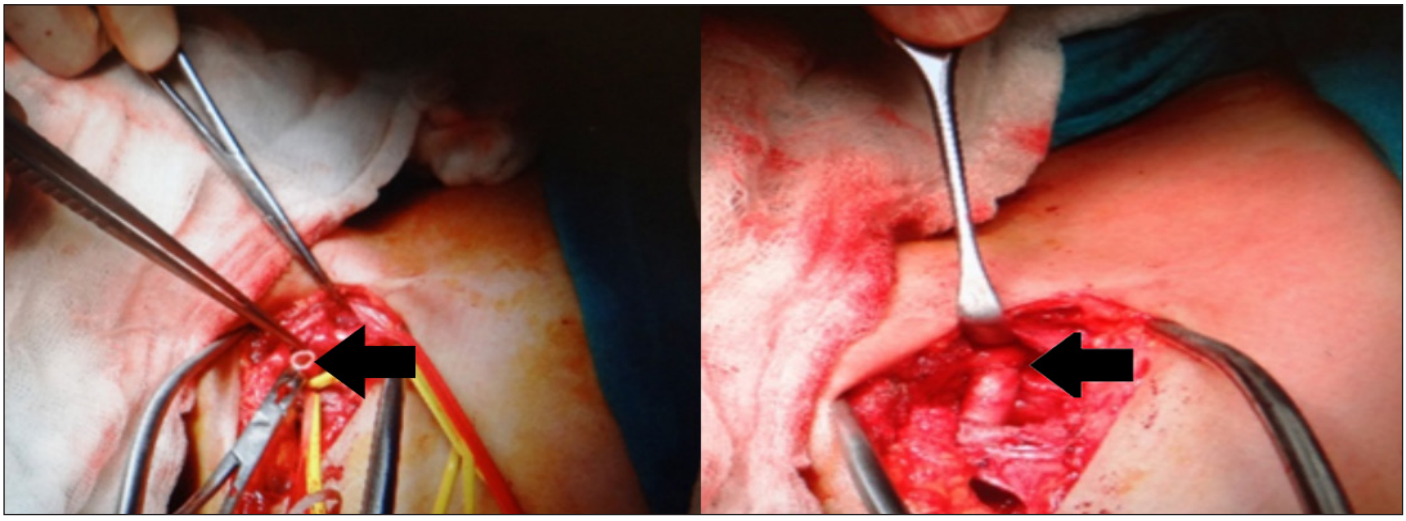

Figure 4. The ARSA (blue arrow points out)

Dysphagia may occur because of many underlying conditions in children as cricopharyngeal achalasia, esophageal achalasia, surgeries carried out because of esophageal atresia (EA) or esophageal stenosis due to the conditions as epidermolysis bullosa, radiotherapy, esophageal polyps or webs (11-15). Among all these diseases, ARSA may be skipped. Contrast studies and esophagoscopy may help for differential diagnosis for these conditions. If there is a strong suspect of ARSA, angio-computed tomography may be one of the best options to certain the diagnosis as it was done in present study.

When the phenomenon is symptomatic, the most common symptoms are dysphagia, respiratory symptoms, chest pain, postprandial bloating, coughing or Horner's Syndrome (7). Treatment choice is depend on the severity of the symptoms. Children with mild or moderate symptoms may be treated with lifestyle and dietary changes whereas patients with severe symptoms may be thought as candidates to surgery (16).

There are considerable amount of surgical methods as median sternotomy, left and right thoracotomies and supraclavicular incision and unfortunately no consensus on which is superior (5).

Atay et al.(5) performed right anterolateral thoracotomy and right supraclavicular incision for three children and one adult respectively. It is mentioned in the literature that since thoracotomy provides better exposure, it should be the choice of approach (3). But also there are studies that advocate supraclavicular incision also provide good exposure and rapid recovery and avoid major interventions as thoracotomy and sternotomy (8). In the three cases in present study, the authors did not experience any difficulties of exposure. Supraclavicular approach also seems to be more minimally invasive than thoracotomy and sternotomy. Also these methods are always on the table and if the one has difficulties of exposure, should not hesitate to convert to thoracotomy.

CONCLUSIONS

It may be suggested that end to side anastomosis of ARSA to carotid artery via supraclavicular incision is a feasible and minimal invasive method for children with dysphagia lusoria.

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