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CASE REPORT

Dysphagia lusoria due to aberrant subclavian artery in a 6-year-old patient: case report

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Abstract

Dysphagia lusoria is a rare congenital condition caused by extrinsic compression of the esophagus by an aberrant right subclavian artery (ARSA), an anatomical variation present in up to 1.8% of the population. The ARSA originates below the left subclavian artery and courses to the right, crossing posteriorly to the esophagus in up to 80% of cases. Although most patients are asymptomatic, symptoms can occur in up to 33% of cases, more commonly in adults and rarely in children. In pediatric patients, early identification is crucial to prevent complications such as difficulty in weight gain, gastroesophageal reflux disease (GERD), and aspiration pneumonia.

The diagnosis is confirmed through imaging studies such as esophagography, CT scans, and magnetic resonance imaging (MRI). In the case described, a 6-year-old patient was diagnosed with dysphagia after experiencing difficulty swallowing solid foods and a significant choking episode. Exams revealed GERD and esophageal compression caused by the ARSA.

Management of dysphagia lusoria can be conservative or require surgery, depending on the severity of symptoms. In this case, initial treatment was conservative, opting for medication to control GERD and the use of prokinetics, resulting in complete symptom improvement after three months of follow-up.

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INTRODUCTION

Dysphagia lusoria is a rare congenital condition initially described as resulting from extrinsic compression of the esophagus by an aberrant right subclavian artery (ARSA)[1,2].

ARSA is the most common congenital anatomical variation of the aortic arch, occurring in 0.4% to 1.8% of the population[1,2]. In this condition, the vessel originates from the left side of the aortic arch, distal to the left subclavian artery, and courses to the right posterior to the esophagus in up to 80% of cases[1,2].

Although most patients are asymptomatic, symptoms may be present in up to 33% of cases[1,2]. Manifestations usually arise mainly in the fourth decade of life, and symptoms at early ages are uncommon. In the pediatric age group, the most common symptoms involve respiratory manifestations, such as stridor and recurrent pneumonia, due to varying degrees of tracheomalacia[2]. As the patient grows, increased thickness and rigidity of the esophageal wall favor the development of dysphagia per se[2]. Indirect signs of this condition in children include weight loss or difficulty gaining weight, prolonged mealtimes, and reduced acceptance of previously tolerated foods[1].

Therefore, early identification of the etiology of dysphagia is important to avoid complications such as feeding disorder/refusal, poor weight gain, gastroesophageal reflux disease (GERD), and aspiration pneumonia[1]. Diagnosis is based on clinical presentation in association with imaging tests such as esophagography and magnetic resonance imaging (MRI)[1].

The aim of this study is to report the case of a pediatric patient with dysphagia lusoria based on information obtained from outpatient assessments and medical record review. This case report was approved by the service's Research Ethics Committee under CAAE number 87734125.7.0000.5639.

CASE REPORT

LSS, a 6-year-old female, sought care with a history of dysphagia to solid foods for approximately 6 months, tolerating pureed and liquid diets without difficulty. There were no prior reports of dysphagia, weight loss, vomiting, or respiratory symptoms. About one month before presenting to our service, she was seen urgently at an outside facility due to choking and food impaction with a piece of apple, which resolved without the need for invasive procedures. Her past medical history was unremarkable. During the diagnostic workup, the patient underwent imaging studies for diagnostic clarification. A contrast esophagus–stomach–duodenum study (ESD) identified gastroesophageal reflux disease (GERD) and an image suggestive of extrinsic compression in the mid esophagus (Fig. 1). Chest computed tomography with intravenous (IV) contrast showed an aberrant right subclavian artery arising directly from the aorta distal to the origin of the left subclavian artery, with a retroesophageal course (Fig. 2). Finally, the patient underwent upper gastrointestinal endoscopy (EGD), which showed a mild narrowing of the esophageal lumen approximately 16 cm from the upper dental arch (UDA), likely associated with extrinsic compression. Initially, conservative treatment was chosen with oral domperidone (0.25 mL/kg three times daily) and omeprazole (1 mg/kg once daily). Approximately three months after starting the medications, the patient became asymptomatic, with good acceptance of an oral diet including solid foods.

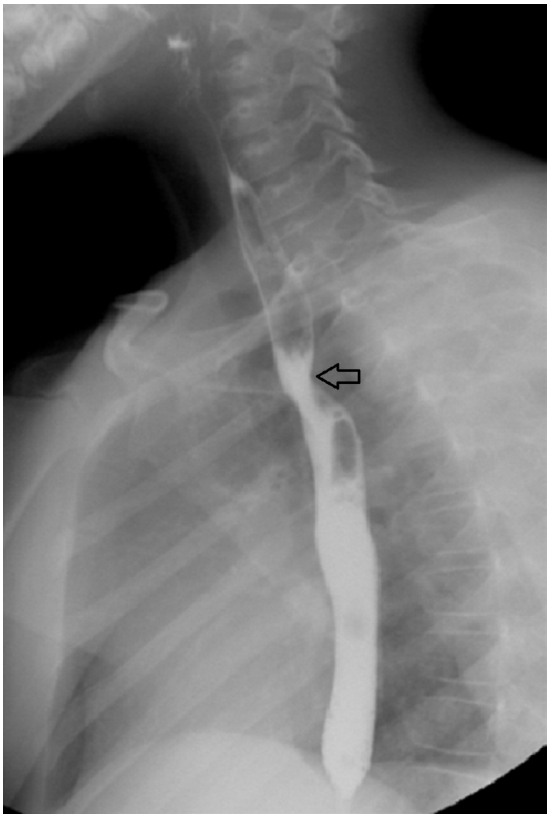


Figure 1. Barium swallow study showing findings suggestive of extrinsic compression of the middle third of the esophagus.



Figure 2. Contrast-enhanced chest CT demonstrated an aberrant right subclavian artery arising directly from the aortic arch distal to the origin of the left subclavian artery, with a retroesophageal course.

DISCUSSION

The most common congenital anomalies of the aortic arch include double aortic arch, anomalous left pulmonary artery, and left aortic arch with ARSA. In addition to being associated with other malformations such as tetralogy of Fallot and pulmonary artery atresia, ARSA has been correlated with up to 36% of fetuses with trisomy 21. Therefore, ARSA should be investigated in every child with Down syndrome and dysphagia[1].

ARSA arises distal to the left subclavian artery and courses to the right posterior to the esophagus in 80% of cases[1]. Although it is rarely implicated as a cause of dysphagia, this condition should be considered in the differential diagnosis in the pediatric age group to prevent potential complications[1].

The diagnostic workup may begin with a barium contrast esophagogram, as it is widely available, does not require sedation, is safe in cases of aspiration, and allows identification of extrinsic compression of the esophagus[1].

MRI can differentiate vascular structures, the trachea, and the esophagus. Computed tomography (CT) should be avoided in children due to radiation exposure; however, it is a useful and effective tool when MRI is not available (as in the present case) or when MRI does not establish the diagnosis with certainty[1].

Therapeutic decisions should be individualized, considering symptom severity and related complications. Asymptomatic patients should not undergo surgical treatment, whereas progressive dysphagia, recurrent aspiration, and significant stridor may be considered indications for surgery[1].

Management of dysphagia lusoria in children may be conservative, including dietary modifications, or may require surgical intervention depending on symptom severity[3,4]. Pharmacological treatment is not well established in the literature and is often used to control symptoms rather than to treat the anatomical abnormality itself[4,5].

Surgical treatment consists of resection of the ARSA and reimplantation via anastomosis to the right carotid artery or the ascending aortic arch; this procedure may be performed via a supraclavicular or thoracic approach and may also involve endovascular techniques[1,2,3,6–8]. Due to the technical difficulty related to arterial vascularization and the artery's diameter, the ideal timing for surgery is when the child is older than 1 year or weighs more than 10 kg[1]. In specialized centers, complication rates are low and long-term outcomes are satisfactory[1].

Treatment of dysphagia lusoria in the pediatric age group should involve a multidisciplinary team including pediatric gastroenterology, nutrition, nursing, and pediatric surgery[1]. In the reported case, the patient was managed with the introduction of a proton pump inhibitor due to GERD associated with dysphagia lusoria, along with a prokinetic agent. She has been followed in the outpatient clinic for 3 months with complete improvement of dysphagia symptoms and good acceptance of an oral diet of any consistency, thus avoiding the need for surgical intervention at this time.

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