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Case Report

Arteria Lusoria: A Rare Cause of Laryngeal Dyspnea in Children: 2 Case Reports

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Abstract

The arteria lusoria is a rare entity, yet it stands as the most prevalent anomaly of the aortic arch. It is usually asymptomatic and rarely revealed in children. Dyspnea, stridor, recurrent pulmonary infections and other symptoms may occur if this vessel pressures the neighbouring structures. Asymptomatic patients undergo medical treatment associated with simple surveillance and surgery is necessary when it becomes symptomatic.

The aim of this article is to show through two case reports in children, the age of diagnosis, the circumstances of discovery, the means of diagnosis and the evolution of this infrequent pathology.

Keywords: *Arteria Lusoria; Laryngeal Dyspnea; Infant; Case Report*

Introduction

The aberrant right subclavian artery or Arteria Lusoria is the most common congenital anomaly of the aortic arch. Its incidence is estimated to be around 0.5 to 2% in the general population. Although typically asymptomatic, this defect can cause dysphagia (dysphagia lusoria), dyspnea, and chronic cough, necessitating surgical intervention. It is unusual for symptoms to develop and progress at an early age. We report two pediatric cases of Arteria Lusoria revealed by laryngeal dyspnea, illustrating the clinical, radiological and therapeutic features of this pathology.

Case Reports

Case 1

Patient information: We report a case of 11-month-old female with a past medical history of premature birth at 36 weeks and neonatal hospitalization for neonatal respiratory distress. The patient has also been followed since a young age for a heart condition known as ventricular septal defect. She is an only child from a 1st-degree consanguineous marriage. The parturition occurred at a hospital facility via vaginal delivery, yielding the birth of a newborn with low birth weight of 2000g. The Apgar score at birth was 9/10/10. The infant was exclusively breastfed until the age of 2 months and was vaccinated according to the national immunization program.

The history of the disease dates back to the age of 3 months, with the onset of recurrent episodes of respiratory distress associated with a hacking cough and laryngeal stridor that worsens during feeds. These episodes of laryngeal dyspnea appeared to respond to nebulizations of adrenaline and to corticosteroids.

She was partially breastfed every 2 to 3 hours and only ate 40-60 mL per feed. Regular cow's milk-based infant formula was used as a supplement. Parents reported that the girl appeared to have difficulty swallowing. They did not notice other symptoms such as vomiting or choking with feeds. She passed normal stool twice a day.

Clinical findings: The infant had no dysmorphic facial features, she was pale, apyretic, weight = 6000g (-3DS), height = 65 cm (-1DS), cerebral palsy (CP) = 37 cm (M), BP = 102/64 mmgh, HR = 120 bpm, FR = 66c/min, cardiac resynchronization therapy (CRT) < 3sec, SaO_2 = 94% on room air. Signs of respiratory struggle such as supra sternal subcostal and intercostal pulling. Cardiac auscultation reveals a holosystolic wheel-radius murmur. Pulmonary auscultation did not reveal the presence of sibilant rales.

Diagnostic approach: The chest X-ray did not reveal any abnormalities. The transthoracic echocardiography showed the presence of a 3 mm interventricular septal defect with no hemodynamic repercussions. Direct laryngoscopy revealed no congenital anomaly of the larynx. A thoracic angioscan, revealed an abnormal vascular structure, which was arising from the aortic arch and then displacing the oesophagus from the back (Figure 1 and 2).

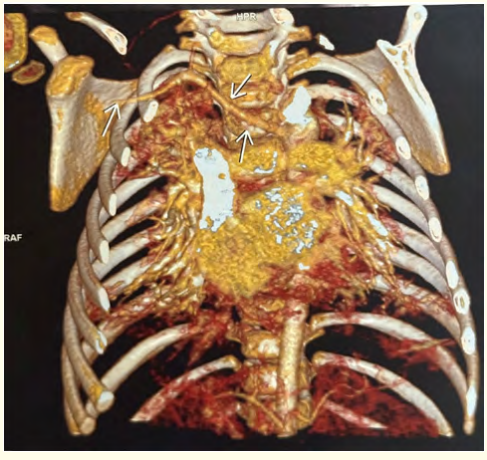

Figure 1: *Coronal image of an angioscan showing the aberrant right subclavian artery passing posterior to the esophagus and trachea at the level of the third thoracic vertebra (Case 1).*

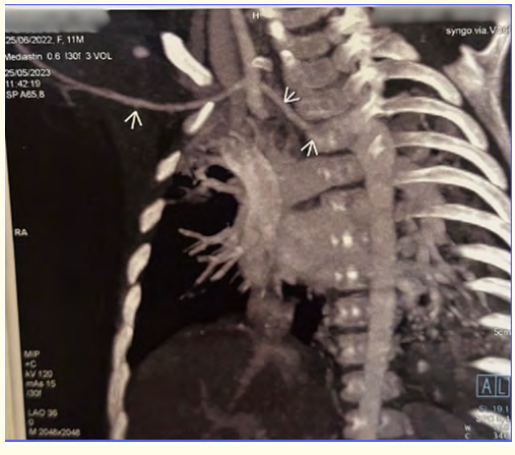

Figure 2: *Aberrant right subclavian artery diagnosed by Angioscan in a 11 old month infant (Case 1). Sagittal image showing the aberrant course of Arteria Lusoria (Case 1).*

Therapeutic intervention: The surgical correction was performed through a right posterolateral thoracotomy. The aberrant right subclavian artery was interrupted. Then, an end to-side anastomosis to the right carotid artery was performed.

Patient perspective: The patient had a satisfactory outcome, with no postoperative complications. Our patient reported complete resolution of their symptoms. She no longer had dyspnea or stridor. She had gained weight, and at 18 months of age, she reached 9000g.

Informed consent: Written informed consent was obtained from the patient's parents.

Case 2

Patient information: We report a case of a 2-months-old Boy who was admitted to the pediatric pneumology unit because of breathing problems since birth. He was born at 38 weeks of gestational age, with a birth weight of 2900g. At 1 week old, he began to present recurrent episodes of inspiratory dyspnea with asymptomatic intercritical interval, treated with outpatient corticosteroid therapy.

Clinical findings: During his hospitalization, the infant continued to present inspiratory dyspnea with stridor exacerbated by exertion. There was no evidence of cyanosis, coughing or vomiting. Clinical examination revealed no specific pathology except for the low weight and the respiratory distress. He weighed only 3,500g and gained only 600g since birth, knowing that he was breast-fed every 2 to 3 hours and had no neurological problems or hypotonia. In light of the lack of clinical improvement with adrenaline nebulization and corticosteroid therapy, the diagnosis of acute laryngitis was called into question. The hypothesis of a laryngeal malformation pathology was ruled out by direct laryngoscopy.

Diagnostic approach: The two-dimensional echocardiogram showed a patent foramen ovale with no impact on blood flow. The precise location of the aortic arch and its branches was unclear. CT angio revealed an ARSA, originating from the left part of the aortic arch just under the left subclavian artery and then progressing to the right side behind the esophagus (Figure 3).

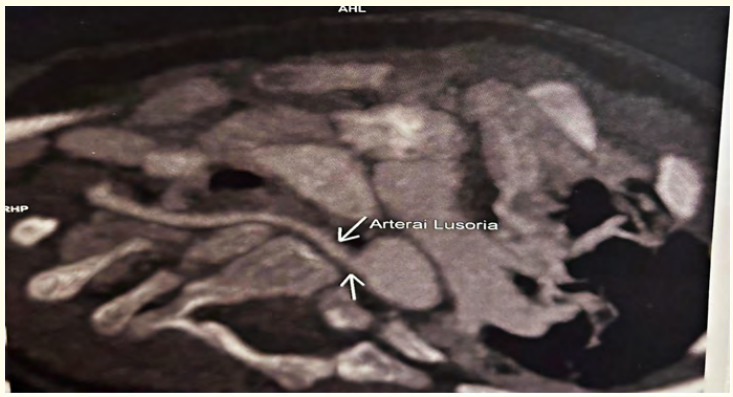

Figure 3: *Transversal slice image of a thoracic angioscan showing the aberrant course of the Arteria Lusoria (Case 2).*

Therapeutic intervention: Intractability and severity of the symptoms made surgical intervention inevitable and acceptable at the age of 4 months. Left anterolateral approach was proceed and the therapy involved a ligation of the RSCA near its origin in the aortic arch and right subclavian-carotid reimplantation.

Patient perspective: postoperative care was provided in the neonatal intensive care unit, he was extubated on awakening and put on corticosteroids and nebulized adrenaline. The evolution was marked by death following a nosocomial infection at D15 postoperatively.

Informed consent: Written informed consent was obtained from the patient's parents.

Discussion

The aberrant right subclavian artery (ARSA) arising from the left part of the aortic arch is a rare congenital anomaly but the most common aortic arch defect. In fact, the aberrant right subclavian artery or right subclavian retroesophageal artery (RSRA) occurs in 0.5% to 2.5% of cases in the general population [1]. The "arteria lusoria" is the name given to it (AL). It originates on the left and extends beyond the left subclavian artery before ascending and turning right toward the right supraclavicular fossa. It passes posterior to the esophagus in more than 80% of cases; it is anterior to the trachea in 5% of cases, and it lies between the esophagus and trachea in 15% of cases [2]. Our two patients had retroesophageal Arteria Lusoria, which is consistent with most cases described in the literature.

Although Hanauld was the first to report this artery anomaly in 1735 from autopsy studies, Bayford was the first to describe the symptomatic form of the condition in 1787, which he called "dysphagia lusoria" or the "freak of nature" in dysphagia patients. Bayford's insight came from examining the autopsy findings of a 33-year-old woman who had passed away due to malnutrition secondary to dysphagia [3].

The ARSA is usually asymptomatic (60 - 80% cases). And While the majority of individuals typically exhibit symptoms around the age of 50, some may show signs of the condition during childhood or even at birth. Symptoms, when present, occur at the two extremes of life. The mean age of the onset of symptoms is 49.9 ± 19.4 years in a systematic study of 141 reports by Michal Polguj., *et al*. The occurrence of arteria lusoria is more common in female than male subjects (55.3% versus 44.7%) in the same study [4].

Patients with an anomalous right subclavian artery (ARSA) may have associated cardiac anomalies such as tetralogy of Fallot, pulmonary atresia, and large aorticopulmonary collateral arteries. In a study, a prenatal testing demonstrates a link between trisomy 21 and ARSA, with 19% to 36% of fetuses having this vascular defect [5].

Due to a lack of rigidity in the trachea, children typically experience respiratory symptoms such as difficulty breathing, chronic cough, wheezing, noisy breathing (stridor), recurring pneumonia, and cyanosis. Van Son., *et al.* found that 86% of infant patients with AL had symptoms of stridor or recurrent respiratory infections [6,7].

In the other hand, the most common presenting complaint in adults is difficulty swallowing (dysphagia), primarily due to anatomical changes that may occur with the aging process. However, dysphagia Lusoria is not specific to adults, and can be seen in children at different ages. This possibility should be kept in mind in the differential diagnosis of dysphagia in children [8].

Chest radiography is typically used as the first step in evaluating respiratory symptoms. The best initial diagnosis would be a barium swallow who may reveal oblique compression of the esophagus between the third and fourth thoracic vertebrae, indicating ARSA. CT or MRI angiography is the gold standard for the diagnostic, replacing traditional angiography: It allows to search for extrinsic compression and to characterize the defective artery. According to Yang., *et al*. the sensitivity of the thoracic CT for the diagnosis of arteria lusoria is 100% [8]. Echocardiography provides a comprehensive assessment of the heart's anatomy and function. If stridor, inspiratory dyspnea or hoarse cough are present, especially before the age of 6 months, flexible bronchoscopy can help rule out a malformative origin and sometimes show the coexistence of tracheomalacia.

Various approaches have been used in the past and continue to be used now to address this condition surgically. The surgical approach involves the interruption and removal of the abnormal right subclavian artery (ARSA), followed by the reattachment of its origin through an end-to-side connection either to the right carotid artery or the ascending aorta, all performed without the need for extracorporeal circulation. When determining the best time for surgery, it is critical to consider the patient's age and weight. One must also consider the

size and diameter of the descending aorta. If the arteries have larger diameters and the child is older, the surgical repair is less difficult and the risk of complications is significantly reduced. The best time for children to undergo surgical treatment is after one year of age or when they weigh more than 10 kg. Complication rates are low (<0.5%) in experienced surgical centers, with excellent long-term outcomes [9,10].

Conclusion

In conclusion, though it is rare, arteria lusoria can appear at a very early age, therefore it is important to consider arteria lusoria an option in young patients with laryngeal dyspnea. No standardized treatment has been determined, though several surgical and non-surgical options have been proposed. The surgical treatment is safe and leads to dramatic relief of symptoms. Progress is often favourable following early and appropriate treatment.

Competing Interests

The authors declare no competing interest.

Authors' Contributions

Nour Al Houda Hajji and Sara Aminou: Bibliography, drafting the article.

Achraf Saidi: Bibliography and Data collection.

Chafiq Mahraoui: Supervision and correction of the article.

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