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Single Incision One Stage Surgical Management of Dysphasia Lusoria - A Case Report

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Abstract: Aberrant right subclavian artery, or arteria lusoria, is a rare congenital anomaly of the aortic arch and may present with dysphagia or regurgitation in infants. Surgical approach remains debatable. Surgical interventions include supraclavicular, thoracotomy approaches and hybrid endovascular approaches with significant limitations and varying degrees of success. This case report describes a median upper partial sternotomy approach whereby the arteria lusoria is transected and reimplanted via a single incision to the right subclavian artery with good excellent outcome and immediate resolution of symptoms.

Keywords: Aberrant right subclavian artery, Dysphasia lusoria, Single incision, median sternotomy approach

1. Introduction

Dysphasialusoria or lususnaturae (freak of nature) was first described by Bayford in 1794.11t is a rare entity caused due to extrinsic compression of the esophagus from any vascular anomaly of the aortic arch. The most common vascular abnormality of the aortic arch is ARSA (Aberrant right subclavian artery), with an incidence of 0.5% - 1.8%.2Surgical treatment remains debatable with several different approaches proposed like supraclavicular and thoracotomy approaches, hybrid endovascular techniques each having their own sets of limitations and success rates. Here, we describe first case of dysphasia lusoria in a six - month old child who was treated using upper partial sternotomy approach.

2. Case

A 6 Month old male child, presented with history of recurrent regurgitation of ingested milk since the age of 40 days of life. Regurgitation occurred immediately or within minutes after every feed. Content was uncurdled milk.

Patient also had history of failure to thrive since the age of 1.5 months. Both regurgitation and failure to thrive improved slightly after introduction of formula feed. There was no history of cough, choking, recurrent pneumonia.

Physical Examination revealed delay in gross motor milestones. Length and weight for age was less than 3rd centile according to the WHO growth charts. There was no significant per abdominal findings.

Initial investigations including complete blood count, serum electrolytes and ultrasound whole abdomen was normal. Gastrograffin meal follow through showed posterior indentation of the oesophagus at D4 level due to external compression suggestive of dysphagia lusoria (Fig 1). It was followed by CECT thorax and CT Angiography, which showed a left sided aortic arch with aberrant origin of the right subclavian artery distal to the origin of the left subclavian artery as the last branch of the arch of aorta (Fig 2). ARSA while crossing the midline to supply the right arm, was causing posterior compression on the oesophagus, thus causing the symptoms.

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Figure 1: Gastrograffin meal follow through showing posterior indentation of the oesophagus at D4 level

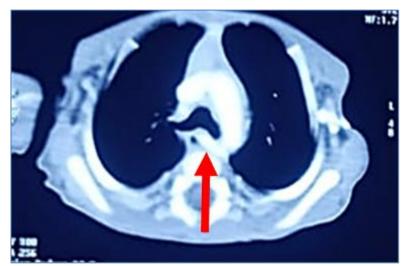


Figure 2: Abberant origin of Right Subclaviasn artery as last branch from the arch

With the diagnosis of dysphagia lusoria due to aberrant Right Subclavian artery the patient was planned for transection and re - implantation of the ARSA to the right common carotid artery using a upper partial medial sternotomy.

Under general anesthesia, in supine position under aseptic precaution skin incision was made from sternal notch to xiphisternum. Upper partial midline sternotomy done followed by right thymectomy. Innominate vein was identified, looped and retracted inferiorly. Aortic arch was left sided and no PDA present. Branches of arch of aorta was identified above the innominate vein. First, Right common carotid artery was identified arising as the first branch from the arch. Right CCA was looped and retracted towards left. Distal right Subclavian artery was identified in the right side in cervical area along which dissection was done till the midline till it was passing behind the esophagus on the right side. The length of dissected ARSA appeared adequate to reanastomose to Right Common carotid artery. So, it was ligated and divided on right side of the oesophagus. The pericardium over the arch of aorta was opened between two stay sutures and aortic arch was retracted towards left and anteriorly. Dissection plane was created between left of esophagus and arch and between left of esophagus and descending thoracic aorta (DTA). Proximal ARSA stump was looped and pulled anteriorly left to the oesophagus where it was again ligated at the origin from the DTA. The distaltransected end of the right subclavian artery was then reimplanted in an end to side fashion to the right common carotid artery using prolene 7 - 0. Pericardium over the aorta was closed. Arterial pulsations was checked in the left brachial artery which was palpable. Sternum was closed with no 2 ethibond, after maintaining haemostasis and placement of one retrosternal drain. Incision site was closed in layers. Patient was started on oral feeds on post operative day 2 and discharged on day 3 of surgery. On follow - up child was accepting feeds and there was no more regurgitation after the surgery.

3. Discussion

Aberrant Right subclavian artery arises most commonly as the last branch from the arch of aorta distal to the origin of left subclavian artery. The most common course is posterior to the esophagus (80%), followed by between the esophagus and trachea (15%) and anterior to the trachea (5%).3Embryologically, it is the persistent 7th intersegmental artery which remain attached to the descending aorta.

Most cases of this anomaly remain asymptomatic and do not require any treatment.

However, rarely patients may present with symptoms of dysphasia (aka dysphasia lusoria), stridor, dyspnoea orchronic cough.

The first successful repair of this anomaly was reported by Robert Gross in 1946 who simply ligated the ARSA by a left thoracotomy approach⁴. Several reports after that proposed

that simple ligation may lead to arm weakness, ischemia and subclavian steal syndrome.

Reconstitution of the divided ARSA was first performed by Bailey and associates in 1965.5 They re - anastomosed the divided right subclavian artery to the ascending aorta. Cooley was the first to reanastomose the distal subclavian artery to the right common carotid artery.6

Nelson et al. in 2020 reported 2 cases of dysphasia lusoria managed by 2 - site operation whereby the arteria lusoria was transected via a left thoracotomy and subsequently re - implanted into the right common carotid artery via a median sternotomy with complete resolution of symptoms.7 The main disadvantage here was requirement of two different incisions which increases the post - operative morbidity.

Dawazah et al. in 2015 published a report "Non aneurysmal aberrant right subclavian artery causing dysphagia in a young girl: challenges encountered using supraclavicular approach" where there was thoracic duct injury during the dissection.8

Isolated upper midline sternotomy approach has not been reported in literature so far. Here, we used an upper partial sternotomy. The advantage of this approach was a single incision. Second, this approach is well versed to cardiothoracic surgeons therefore identification of structures is easier compared to the left thoracotomy or supraclavicular approach where sometimes it is difficult to identify the structures and less chances of injury to the structures like Recurrent larvngeal nerve which may be injured with a supraclavicular approach as usually along with ARSA there is a non - recurrent course of the right recurrent laryngeal nerve. Third the origin of the ARSA was easy to reach as the arterial stump was looped and pulled out anteriorly and ligated close to the origin. Thus, recurrent symptoms due to large Kommerell's diverticulum left in supraclavicular approach can be avoided. Reimplantation of the distal subclavian artery to the right common carotid artery is also easier through this approach. Also, this approach gives choices for reimplantation viz ascending aorta, arch of aorta, right common carotid etc.

Thus, upper partial median sternotomy is a feasible approach with less complications and complete resolution of symptoms.

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