Incidentally diagnosed dysphagia lusoria in a case of bladder carcinoma

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Abstract

A retro esophageal right subclavian artery, the most common congenital aortic arch abnormality, is an unusual cause of dysphagia in adults. The embryologic abnormality of the aortic arch involves involution of the fourth vascular arch, along with the right dorsal aorta, leaving the seventh intersegmental artery attached to the descending aorta. This persistent intersegmental artery assumes a retro esophageal position as it leaves the thorax into the arm. Since compression of the esophagus by this right subclavian artery may produce dysphagia, the term "Dysphagia Lusoria" has been used to describe the symptom complex. Here we present a case of a patient with incidentally diagnosed Dysphagia Lusoria.

Key Words: Dysphagia Lusoria, Aberrant right subclavian artery.

Introduction

The Aberrant Right Subclavian Artery (ARSA), also known as "Arteria Lusoria" (AL), is the most common of the intra-thoracic embryologic anomalies involving main arteries, with an incidence of 0.4% to 2% [1]. Most of the patients presenting with this anomaly are asymptomatic. However, its presentation may vary from mild to moderate, and sometimes, severe dysphagia. In children it may present with stridor and recurrent chest infections.

Case report

A 65 year old male presented with complaints of burning micturation and intermittent haematuria since 6 months. There were no GI or respiratory symptoms. On general examination it was found that the blood pressure recorded in both the upper limbs was different, a fact noticed during the pre anaesthetic check up prior to the proposed diagnostic cystoscopy for the haematuria . The blood pressure was within normal limits in the left upper limb(120/76 mm of Hg), while in the right upper limb it was persistently high during all readings(160/100mm of Hg) in the supine position. All routine investigations including electrocardiogram and chest X ray were normal including a 2 D Echo cardiogram with an ejection fraction of 55%. A cardiology opinion taken suggested a CT angiography to rule out causes for the discrepancy in blood pressure recordings, mainly co-arctation of aorta. CT angiogram revealed presence of an aberrant right subclavian artery, which was partly thrombosed with multiple collaterals in the surrounding areas(Fig 1). Partial compression of the oesophagus was seen with the aberrant artery pushing it anteriorly. This finding prompted an upper GI Endoscopy and Barium swallow which again were normal (Fig 2). Meanwhile a CT Urogram done for his presenting complaint of haematuria was suggestive of a bladder mass for which Trans Urethral Resection of the Bladder Tumor (TURBT) was planned. As the patient was essentially asymptomatic, cardiology fitness was

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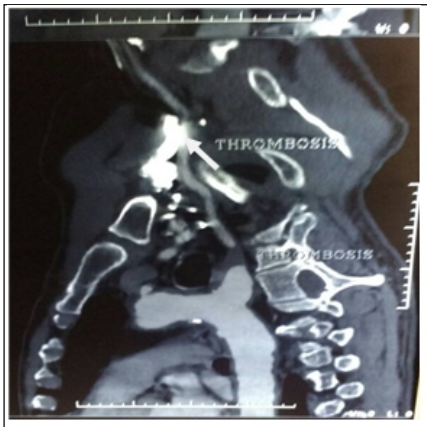

Figure 1: CT Angiogram showing abnormal Right Subclavian artery with thrombosis

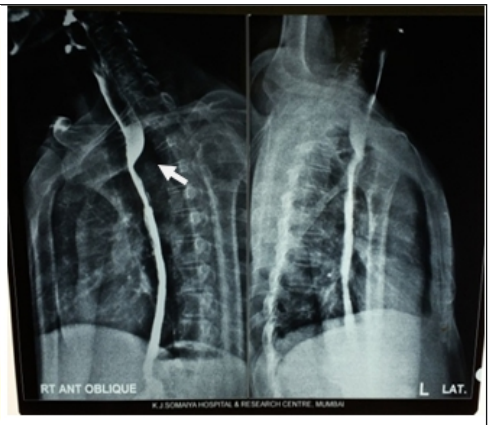

Figure 2: Barium swallow showing a normal esophagus

obtained and a TURBT performed. Post operative recovery was uneventful. The patient was started on Aspirin and Clopidogrel prior to discharge.

Discussion

This anomaly first reported in 1794 by London physician David Bayford, was originally described as "dysphagia by freak of nature," and is now commonly referred to as dysphagia lusoria [2]. Burckhard Komerell is credited with the first radiological description, in 1936, as the condition had only been diagnosed at postmortem prior to that time. He stated that the pulsating mass behind the oesophagus does not consist of the right subclavian itself, because the calibre of this vessel is much smaller and felt that it was more likely that this mass consists of an aortic diverticulum, from which the right subclavian artery originates [3]. The most common embryologic abnormality of the aortic arch is an aberrant right subclavian artery, which occurs in 0.5% to 1.8% of the population [4]. The abnormal right subclavian artery origin can be explained by the involution of the 4th vascular arch with the right dorsal aorta. The 7th intersegmental artery remains attached to the descending aorta, and this persistent intersegmental artery becomes the right subclavian artery. This leads to the formation of an aberrant artery, which often occurs posterior to the oesophagus [5]. Most of the patients presenting with this anomaly are asymptomatic. However, its presentation may vary from mild to moderate and even severe dysphagia. In children it may present with stridor and recurrent chest infections. The difference in presentation between pediatric and adult age groups seems to be related to the absence of tracheal rigidity in children and the development of physiologic and anatomic changes such as increased stiffness of esophageal and vessel walls with increasing age [6]. Symptoms usually occur when a ring of vessels forms around the esophagus causing extrinsic compression leading to dysphagia. The 1st successful repair of this anomaly was reported by Robert Gross in 1946 [7]. He described dividing and ligating the ARSA via a left thoracotomy, in a 4 month old infant. Lichter was the first to describe surgery on an adult with this condition in 1963. Early reports revealed that simple division without restoration of flow leads to weakness and ischemia of the right arm, which can cause a reversal of blood flow from the right vertebral artery to the right subclavian artery. This phenomenon, first described by Contorni in 1960 [8], was named 'subclavian steal' by Reivich and colleagues in 1961 [9]. In 1964, Hallman and Cooley recommended an arteriogram prior to surgical repair of the congenital aortic vascular ring in adults [8,9] Conventionally the treatment consisted of proximal ligation of the aberrant right subclavian vessel. However, it was found to result in Subclavian steal phenomenon, hence it was not preferred [8,9]. It is not until the last thirty years that surgery has become the standard therapy for this condition, and several authors have advocated various approaches. Pome et al in a review of the literature published in 1987, found only twenty reported surgically treated patients. Janssen et al reported six cases of dysphagia lusoria diagnosed and managed between 1992 and 1997. Three patients responded to either medical management like antacids, or dietary modification. One patient underwent a right carotid-subclavian end to side bypass via a right supraclavicular approach. Two patients underwent a two incision approach (i.e., thoracotomy and cervical incision) [10]. The standard operative treatment now involves proximal ligation of aberrant subclavian artery and establishment of flow by means of anastomosis of distal subclavian to right common carotid artery. The right supraclavicular approach was found to provide excellent exposure for proximal ligation and distal anastomosis of the aberrant right subclavian artery to the right common carotid artery in adults, while in children a right anterolateral thoracotomy in the 4th intercostal space by muscle sparing technique is preferred. In both adults as well as children care is taken to preserve the major structures such as the brachial plexus, right Vagus and right Recurrent laryngeal nerve. Distal portion is anastomosed with right carotid artery [11]. AD Rogers and others described the various strategies in management of aberrant right subclavian artery [12]. Surgical access included a left or right thoracotomy/ median sternotomy or trans-sternal, approach. A cervical, right supraclavicular, or endovascular and hybrid approach was also suggested. Reconstruction methods included simple ligation and division, right subclavian artery to ascending arch anastomosis, right subclavian artery to carotid artery anastomosis or stent-grafting for patients with concomitant aneurysmal disease. Few cases have been reported to have been managed by endovascular or hybrid approaches. Although long-term results are awaited, initial results are promising. Shennib and Diethrich have described minimally invasive, hybrid endovascular approaches. A right supraclavicular approach was used to perform a right carotid-subclavian bypass prior to division of the right SCA. They then deployed an occluder in an antegrade fashion into the proximal end of the right SCA in order to maintain good control of the deployment and to avoid embolization of the occluder into the aortic arch from a retrograde approach. A right femoral artery access was

used via a 9F sheath [13].

Conclusion

Dysphagia lusoria is a rare clinical entity and even more rarely presents with symptoms. It is usually incidentally diagnosed while patient is being investigated for some other medical condition. Diagnosis is made by CT Angiogram which shows aberrant origin of right subclavian along with retroesophageal right subclavian artery. Management is reserved only for symptomatic cases and is surgical.

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