

A Rare Case from Eastern part of Nepal: Young Female with Right-Sided Bovine Aortic Arch with Coarctation of Aorta and Takayasu's Arteritis

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ABSTRACT

'Bovine' a misnomer term refers to a group of congenital abnormalities of the aorta in which there is aberrant origin of the left common carotid artery. Takayasu's arteritis is a rare, chronic inflammatory disease affecting the walls of the aorta and its main branches leading to stenosis, occlusions, dilatation, and aneurysms of involved vessels. It is rare to find both in a single patient. We present a 20 year young Muslim female from eastern Nepal complaining of pain and weakness of all four limbs, with cold digits for 3 months.

Keywords

Aorta surgery; bovine aortic arch; coarctation of aorta, rheumatology; Takayasu's arteritis

INTRODUCTION

Bovine, a misnomer term refers to a group of congenital abnormalities of the aorta in which there is aberrant origin of the left common carotid artery.¹ Coarctation of aorta among all congenital heart diseases accounts for 6-8% .² Takayasu's arteritis as described by Mikito Takayasu, known as "Pulseless disease" is common in Asia and the Far East.^{3,4} Inflammatory cell infiltration found in Takayasu's arteritis suggests autoimmune pathogenesis commonly involving the aorta and its branches leads to stenosis, occlusions, dilatation, and aneurysms.⁵ To our knowledge, this case of Takayasu's arteritis and right-sided bovine aortic arch with coarctation in the same patient is the first ever described.

CASE PRESENTATION

A 20-year-old female, known case of Takayasu's arteritis and on oral Methotrexate, presented with complaints of bilateral upper and lower limb pain and weakness for 3 months, associated with cold digits. On

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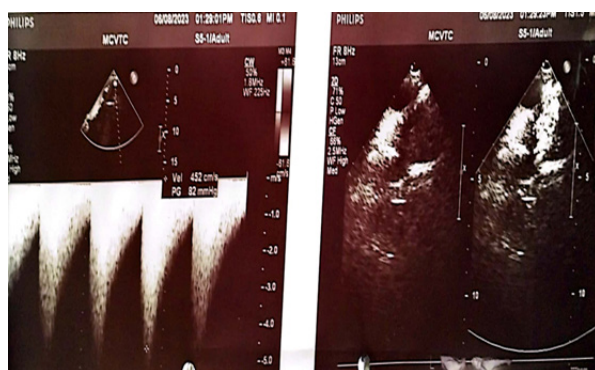


Figure 1. Showing peak gradient across the Coarctation segment 82 mmhg with flow velocity of 452cm/s with pan-diastolic spill

examination, Blood Pressure on the left and right upper and lower limbs was 118/90 mmhg (Millimeter of mercury), 144/98mmhg, 120/90mmhg, and 120/88mmhg respectively. The patient had cold bilateral hand and foot with increased sweating and feeble pulse in the groin and feet along with carotid bruit on auscultation.

Echocardiography (Fig. 1) revealed, severe narrowing of aortic arch- post ductal coarctation of aorta with peak gradient 82mmhg with a pan-diastolic spill. Computed Tomography Aortogram (Fig. 2) showed Right-sided aortic arch with aberrant origin of left subclavian artery, a suspicious fibrous sling between left subclavian artery and left pulmonary artery.

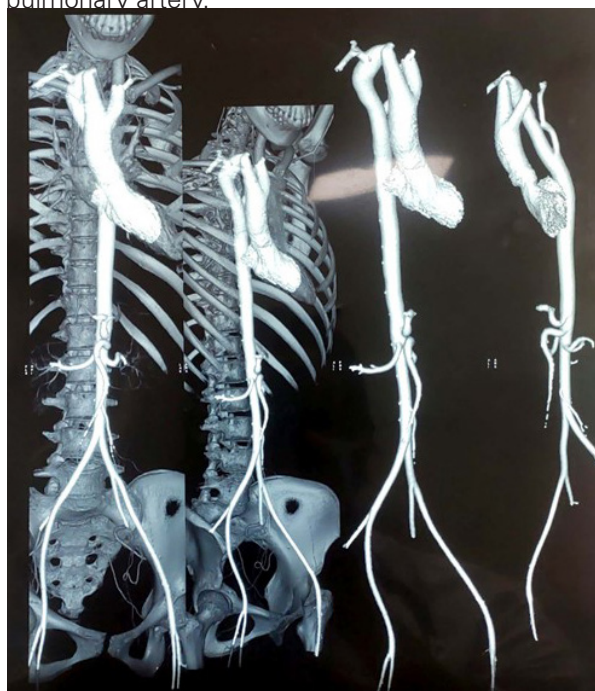


Figure 2. Computed Tomography Aortogram showing Bovine type Right-sided Aortic Arch with Coarctation segment high up about Left Clavicle

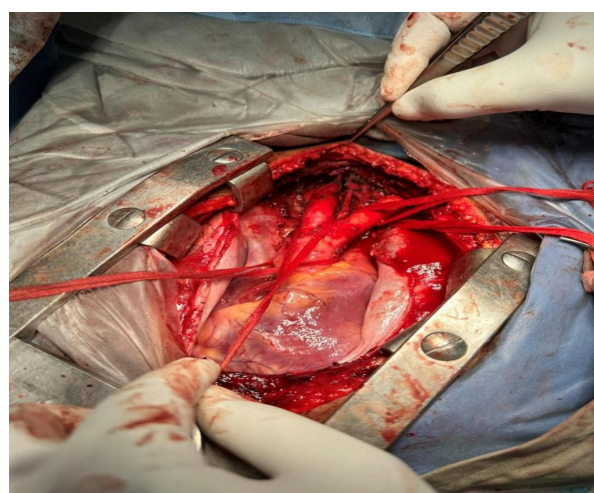


Figure 3. Dissecting out bovine arch along with its innominate branches

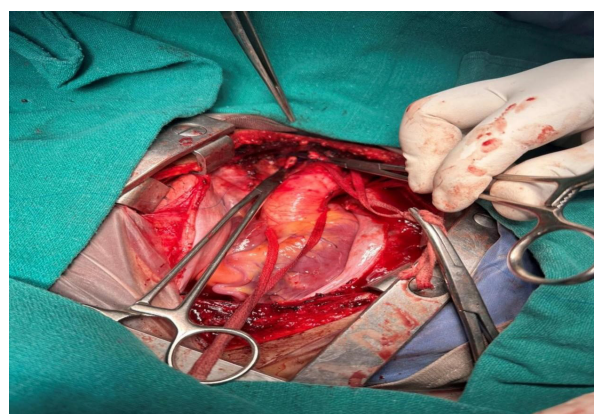


Figure 4. Limited space for clamping and anastomosis of the proximal and distal parts of the Aorta after coarctation segment resection

Rheumatology review was done and advised for Methotrexate to be kept on hold for 1 week before any planned surgical intervention. Coarctation repair was performed via Standard median sternotomy incision with dissection of the extra pericardial part of the aorta. The aorta was freed along with its innominate branches and taken control by umbilical tapes as shown in (Fig. 3) below. Intra-operative findings was noted which showed a Right aortic arch with the aberrant origin of left subclavian artery, fibrous sling between left SCA and left PA, severe stenosis at the level of Arch between two sub aortic vessels, and left Brachiocephalic Vessel post to the aorta. The intra-operative challenge presented was to clamp the aorta proximal and distal to coarctation in the limited space (Fig. 4) and to complete the proximal and distal anastomosis end-to-end after mobilization to ascertain tension-free anastomosis. Resection and end-to-end anastomosis was done with 4.0 polypropylene sutures.

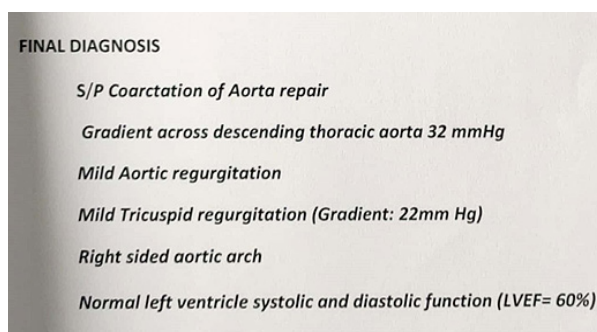


Figure 5. Decreased gradient across the coarctation segment

Post-operative stay in the hospital was uneventful. The patient was discharged and Methotrexate restarted. On 6 month follow-up period patient was asymptomatic with no complications. Her echocardiography results showed decreased gradient across the coarctation segment as shown in the (Fig. 5).

DISCUSSION

The recently revised, American College of Rheumatology/EULAR classification has been developed for the diagnosis of Takayasu Arteritis. The criteria include 2 absolute requirements, seven additional clinical criteria, and 3 additional imaging criteria with a total of 19 points. A patient with a cumulative score of ≥ 5 points meets the requirement. Our case fulfills the absolute criteria with 8 points on the cumulative score fulfilling the diagnostic requirements.⁶

Bovine arch variant was found in 13.6%, with a prevalence of 26.8% in the African populations.⁷ Type I Bovine Arch (T1BA) 14.9%, presents with a common origin of the innominate and left common carotid arteries at the level of aortic arch, and Type II Bovine Arch (T2BA) 16.2% presents with the left common carotid artery originating directly from the innominate artery at an average distance of <1 cm and not greater than 2.5cm. There was a trend toward higher prevalence of both T1BA and T2BA among dissection patients.²

Initially, diagnoses were done with angiography. But today, most centers use electrocardiography for simplifying diagnostic procedure. Spiral computed tomography also helps in diagnosis of aortic arch malformation.⁸

The association between coarctation of the aorta and bovine arch anatomy has been reported in up to 28% of cases with a higher incidence of re coarctation after arch repair in cases with bovine arch anatomy.⁷ In a study, it has been found that only 50% of bovine arch patients were free from adverse effects of re coarctation as compared to 80 % in the general population.⁹

CONCLUSION

The complications of aortic pathology in the Bovine arch compared to the general population, increased risk of re-coarctation, the difference in approach due to varying anatomy and complications added by Takayasu arteritis warrants further studies in this subject. Thus along with aortic pathology, this case study highlights the hindrances surgeons face while operating such cases with a combination of Bovine arch, co-arctation of aorta and Takayasu arteritis.

CONSENT

Written informed consent was taken from the patient for the case report publication.

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CONFLICT OF INTEREST

The author(s) declare that they do not have any conflicts of interest with respect to the research, authorship, and/or publication of this article.

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