AORTO ARTERITIS WITH EXTENSIVE VASCULAR CALCIFICATION IN A MALE WITH PRIMARY **HYPOGONADISM**

Author's Affiliation:

*Professor & Head, **Junior Resident, ***Senior Resident, ****Assistant Professor, ****Associate Professor, *****Additional Professor, Dept. of Medicine, Govt. Medical College, Kozhikode- 673008, Kerala, Trdia.

Corresponding Author: Dr. P. K. Sasidharan,

Professor & Head, Dept. of Medicine, Govt. Medical College, Kozhikode- 673008, Kerala, India

E-mail: sasidharanpk@gmail.com Sasidharan P.K.*, Paul Alappat**, Manuprasad***, Sreejith R.****, Praveen M.****, Geetha P.*****

ABSTRACT

We report the case of a male with Aorto arteritis (Takayasu's) with extensive calcification of aorta and its branches. Extensive calcification of the arteries is unusual in Takayasu's arteritis. A variety of mechanisms may play a role in this phenomenon of calcification, and we attempt to elucidate some of them.

KEYWORDS

Vasculitis; Aortoarteritis; Vascular calcification; Takayasu.

Case Report

A 38 year old male, working in a construction company as a mason for 22 years from Calicut presented with history of insidious onset slowly progressive intermittent claudication in both lower limbs for the last three years. At presentation it has reached a stage that he had to rest every 5 minutes during walking. He married twice but had no children from either marriage. He had consulted for infertility and was found to have a low sperm count. He also had history of abdominal pain following meals suggestive of mesenteric ischemia and history suggestive of migraine with aura for the last 10 years. There was no history of pain in the buttocks or in upper limb; there was no history of impotence, chest pain, palpitation or syncope. He had no history of arthritis, fever, photosensitivity, flank pain, hematuria, jaundice or vomiting. He was a non smoker but used to drink alcohol 200 ml of brandy almost 5 days a week but stopped since the last eight months. His elder brother and sister have no children the cause of which is not known.

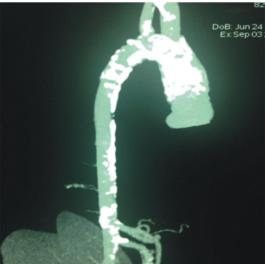
On examination he had sparse facial, axillary, pubic and body hair with gynecomastia and atrophic testes. He had a coarse facies with protruded jaw. Skin and nail changes suggested early chronic liver disease. Pulse rate was 68/mt regular, normal in volume and character, no radiofemoral or radioradial delay. Upper limb pulses were normally palpable, but both femorals were feeble and Popliteal, posterior tibial and dorsalis pedis were absent in both lower limbs. Bruit could be heard over femoral, renal and carotid arteries. Blood pressure in the right upper limb was-120/80mm

Fig 1: MR angiography



Fig 2 & 3: CT Angiogram: Showing extensive calcification of the whole of aorta and its branches





Hg and left upper limb was 116/74mmHg. Cardiovascular system examination revealed an ejection systolic murmur in the aortic area. Other systems were within normal limits. Fundus examination was normal. Gastrointestinal, Respiratory and CNS examination were normal.

Investigations

Hb-11.7g/dL, TLC-7200/mm³ DLC P74 L24 M2, Platelet count was 2.08 lakh/mm³ . Hct-34.8%, ESR-23mm in first hour, Blood sugars, renal function, liver function and electrolytes were normal. Urine did not show any sediments or proteinuria. Serum Calcium was 9.4 mg/dL, Phosphate 3.4mg/dL and Alkalinephosphatase was 129IU. ECG was normal;

Fig 4: X-ray showing vessel calcification



Parathormone level was normal-17.7ng/L(15-65). There was no dyslipidemia and the screening for infections like Tuberculosis HIV, HBsag and HCV were negative. ANA and APLA were also negative in low titers. Serum Cortisol and Thyroid functions were normal. Serum Testosterone was low-0.518ng/ml(2.8-8.0), with high FSH-39.9mIU/ml(1-14) and LH-28.5mIU/ml(0.7-7.4) and normal Prolactin-9.2ng/ml(2.5-17.0). Semen analysis did not show the presence of sperms. Chest X-RAY was normal and the Mantoux test was negative. USG whole abdomen findings & Doppler were unremarkable except, early renal parenchymal disease was echo cardiography done.

MR Angio reported as: showing minimal luminal irregularity but no definite opinion could be given.

MRAngiogram Report

Minimal luminal irregularity involving distal aspect of right superficial femoral artery, Otherwise normal. Since he had definite features of large vessel disease involving both lower limbs and clinically mimicking coarctation of aorta we went ahead with CT angiogram.

Considering possibility of Takayasu only a magnetic resonance angiogram was done but to our surprise it was inconclusive, since the clinical suspicion was so high that we went ahead with CT angiogram, which revealed extensive calcification of ascending aorta, arch, descending aorta, branches, extending till anterior tibial and dorsalis pedis hence confirming our initial clinical diagnosis. Calcification was also noted in the superior mesentric artery-probably the reason for the postprandial abdominal pain. Retrospectively on reviewing the x-ray foot which was taken elsewhere for heel-pad thickness when someone suspected acromegaly, since he had coarse facial feature- it showed calcification of the arteries

DISCUSSION

This middle aged male had typical history and findings of large vessel vasculitis mimicking Takayasus arteritis. The only other disease we wanted to excude was coractation of aorta. Clinically it cannot be atherosclerotic vascular disease since such extensive involvement to the extent to produce calcification and still not having angina and that too with no Dyslipidemia, is unusual. If at all it has to be an inflammatory disorder followed by calcification in the given clinical setting. He also had features of hypogonadism clinicaly which on subsequent investigations was found out to be primary hypogonadism with absent sperms in semen analysis. His siblings also have no children. So we attribute an inherited cause for the hypogonadism. The final diagnosis was thus large vessel vasculitis, Takayasu arteritis with extensive calcification- with primary hypogonadism. This case demonstrates that while mechanisms of vascular calcification are poorly understood, inflammation per se might be sufficient to mediate increased mineral stress leading to vessel calcification, even in the absence of hypercalcmia or hyperphosphatemia or renal impairment. The relation with primary hypogonadism is not previously reported in literature. One study has shown patients with hypogonadism have abnormal calcium metabolim leading to increased bone resorption and increased urinary excretion of calcium³. This may have also contributed to the extensive calcification in his arteries. We could get only three reports from the Literature search on such vascular calcification.

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