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RESEARCH ARTICLE

BALLOON ANGIOPLASTY FOR AN UNUSUAL CAUSE OF BILATERAL BRACHIAL ARTERY OCCLUSION: A RARE MANIFESTATION OF FIBRO MUSCULAR DYSPLASIA

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ABSTRACT

Sub acute or chronic upper limb ischemia is often non-atheromatous, especially in the young and is of multiple etiologies. Subclavian artery is affected the most in atheromatous/non atheromatous etiologies but brachial artery is most affected in fibro muscular dysplasia of upper limbs. Here we have a 58-year-old man presented with right upper limb claudication for the last 1 year and on investigation found to have bilateral brachial artery occlusions was treated successfully with Percutaneous balloon angioplasty. This is a very rare case of bilateral brachial artery occlusion with a classical beaded appearance of the renal arteries giving the only clue for etiology of fibro muscular dysplasia. Excellent response to balloon angioplasty without stenting was highlighted in this case.

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INTRODUCTION

Fibro muscular dysplasia (FMD) is a non atherosclerotic, non inflammatory vascular disease that involves primarily the renal and internal carotid arteries and less often the vertebral, iliac, subclavian, and visceral arteries. Although its pathogenesis is not completely understood, humoral, mechanical, and genetic factors as well as mural ischemia may play a role. The natural history is relatively benign, with progression occurring in only a minority of the patients. In this case we would like to show the unusual presentation of fibro muscular dysplasia which an interventional cardiologist must be aware of while doing a procedure

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CASE PRESENTATION

A 58 year old male hypertensive, non diabetic, presented with rest pain of right forearm and hand for 4 days. He also gave past history of claudication pain of the right forearm and hand on washing clothes since the past one year. He had no prior history of smoking and had no similar symptoms in left hand or lower limbs. On examination, he had feeble brachial and radial artery pulse in both upper limbs. Right upper limb pulse was feebler than the left upper limb. There were no other ischemic signs in both the upper limbs. All the other pulses were normal. Rest of cardiovascular examination was normal. The Doppler evaluation of both upper limb arteries showed normal velocities at both subclavian arteries but decreased velocities in both brachial's. All the initial screening laboratory tests, which included complete blood count, erythrocyte sedimentation rate, antinuclear anti-body, rheumatoid factor, lipid profile etc yielded results in normal ranges Peripheral angiography showed long segment total occlusion of both brachial arteries. The distal brachial arteries were reformed on both sides with contrast filling both radial and ulnar arteries (Figure 1 and 2). Patient was treated with Percutaneous balloon angioplasty with good distal flow achievement (Figure 3-5)

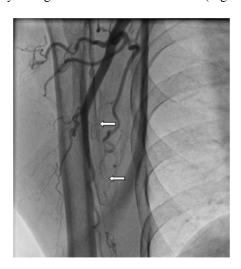


Fig1. Before angioplasty Right brachial artery: note the long tubular/string like lesion continuing with the occluded brachial artery



Fig. 2. Before angioplasty Left brachial artery: note the long string/tubular occluded part of brachial artery



Fig. 3. Post balloon angioplasty of right brachial artery: Note the smooth walled artery after angioplasty



Fig. 4. Balloon angioplasty of brachial artery: Done with Admiral Xtreme Percutaneous Angioplasty balloon (Medtronic)



Fig. 5. Post angioplasty left brachial artery flow: Note the Smooth Arterial wall after the angioplasty

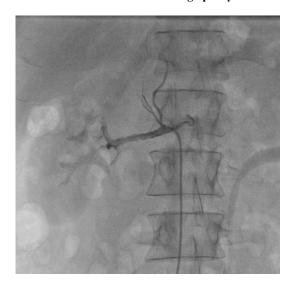


Figure 6. Right Renal angiogram: showing a typical 'beaded appearance' with No luminal obstruction

As the atherosclerotic involvement of the brachial arteries is rare and the pattern of lesion was unusual and no other aetiology was obvious, we did coronary angiography as well as the angiography of the carotid, vertebral and renal angiography. To our surprise, renal angiography showed classical beaded appearance and hence the diagnosis of fibro muscular dysplasia was made (Figure 6)

INVESTIGATIONS

ANA: negative

Mauntaux test: negative

ESR: 21 mm Hb: 15 gm% TLC: normal

Renal function tests: negative

DIFFERENTIAL DIAGNOSIS

Vasculitis/Takayasu arterits Trauma Fibro muscular dysplasia

TREATMENT

Percutaneous Angioplasty of right brachial was performed first via right femoral artery. We placed a 8 F femoral sheath (Cordis®, USA) into the femoral artery and used a 6F MullinsTM (Cook) sheath up to the subclavian artery for a good support. First we tried to enter the lesion with a j tip 0.035 "Terumo wire (Terumo®, Japan), which passed very smoothly. Then the lesion was dilated with a 4 x 40 mm peripheral PTA Balloon Admiral xtreme (Medtronic) at 6 atms for 15 seconds all through and showed a good flow distally. With this result we proceeded for PTA on left brachial artery. This was also a long segment occlusion with a good collateral flow and a good distal brachial artery formation. We followed the same steps using j tip 0.035" Terumo wire and a 6F Mullins™ (Cook) sheath up to the left subclavian artery. Again the lesion was crossed successfully. We did balloon dilatation with the same Admiral xtreme balloon 4 x 40 mm at 6 atms pressure for 15 seconds each time. The end result was good with a brisk arterial flow. No complications occurred during the procedure.

DISCUSSION

Fibro muscular dysplasia (FMD) is a non-inflammatory non-atherosclerotic disease that affects small and medium size arteries (Olin *et al.*, 2012) Woman in their 40s are primarily affected. Renal and carotid arteries are the most commonly involved vascular beds (Mettinger, 1982). Other vascular beds can be affected although less frequently (Mettinger, 1982). They are few case reports of FMD involving the brachial arteries (Kolluri and Ansel, 2004) There are 14 case reports since Kessler reported the first case of FMD in 1982, (Kolluri and Ansel, 2004) FMD involving bilateral brachial arteries has been reported only 5 times thus far in the literature (Suzuki *et al.*, 1999; Cutts and Grewal, 2000; Reilly *et al.*, 1993). The fifth case was reported by Kolluri in 2004. Kincaid *et al.* (1968) proposed an angiographic classification of FMD into four types.

Multifocal type, with multiple stenosis and the 'string-of-beads' appearance, Tubular type, with a long concentric stenosis, Focal type, with a solitary stenosis less than 1 cm in length, and Mixed-type stenosis. Histological FMD affects intima, media, or adventitia and has been classified depending on the vessel wall layer involved. Medial fibroplasia is the most common subtype of this category and accounts for approximately 70-95% of all the FMD cases reported. It is characterized by the typical string of beads appearance as in the renal artery of this patient. Subclavian artery is affected the most in atheromatous as well as non-atheromatous aetiologies but brachial artery is the most affected in fibro muscular dysplasia of upper limbs (Plouin et al., 2007). Management of FMD includes angioplasty of the involved arterial bed as illustrated by our case. Stenting is not indicated for these fibrotic lesions, as recurrence of FMD is rare. There have been several approaches to treating FMD in brachial arteries. Medical management of brachial artery FMD is not effective, (Olson et al., 1984) Resection of the affected portion and reverse vein grafting has been successfully used (Kelly et al., 1982). Angioplasty with or with-out intra arterial thrombolytic therapy has also been reported to have good success (Ciocca et al., 1995). Stenting of the vessel is usually not needed (Kolluri and Ansel, 2004).

LEARNING POINTS

- Fibro muscular dysplasia is a rare disease of upper limb arteries
- Bilateral simultaneous involvement of brachial artery narrowing should raise the suspicion of fibro muscular dysplasia.
- Treatment for even the long tubular form of fibro muscular dysplasia as seen from this case was plain balloon angioplasty. There is no need for stents.
- Recurrence is rare after balloon angioplasty

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