

A rare presentation of Takayasu Arteritis in Sub-Saharan Africa: A case report in a young female from Mauritius

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ABSTRACT

Background

Takayasu's arteritis (TKA) is a severe inflammatory form of large vessel vasculitis that primarily affects the aorta and its major branches. TKA is a rare disease, with a reported worldwide incidence rate of only 1 to 2 cases per million.

Case presentation

This is a case report from Mauritius of a female 33 years in age suffering from TKA. Individuals with TKA exhibit infiltration of inflammatory cells into large vessels, resulting in the release of proinflammatory cytokines, including IFN- γ , TNF- α , IL-6, IL-8, IL-17A and IL-18. Both the clinical assessment and laboratory assessment of TKA are challenging. This instance serves as a poignant reminder of the potential severity and complexity of TKA.

Conclusion

This report highlights and sheds light on the intricate process of diagnosing and managing TKA, accentuating the importance of thorough clinical assessments and meticulous history taking. By diving deep into the patient's medical background, signs and symptoms as well as with the proper investigations, healthcare providers can swiftly recognize the distinctive features of TKA. This report also highlights the long-term follow-up for disease progression and the efficacy of treatment for patients diagnosed with TKA.

Keywords

large vessel vasculitis, rare autoimmune disease, autoimmune rheumatologic diseases, diagnostic reference levels, takayasu arteritis

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Background

Takayasu's arteritis (TKA) is a rare disease, with a reported worldwide incidence rate of only 1 to 2 cases per million, with the majority of cases occurring in females in a ratio of 9:1 [1]. TKA causes severe inflammation of large vessels that primarily affects the aorta and its major branches [2]. In the lesions, multiple immune cells (CD4+ T cells, CD8+ T cells and macrophages) secrete various cytokines which cause injury to the involved arteries [3]. Inflammatory cells infiltrate large vessels, releasing proinflammatory cytokines and acute phase reactants viz. (CRP) [4]. The clinical and laboratory assessment of the disease is challenging. To diagnose TKA, the new classification criteria was published in 2022 by the American college of Rheumatology (ACR) and the European Alliance of Associations for Rheumatology (EULAR) [5]. A score of ≥ 5 is suggestive of TKA. In respect to treatment, TNF inhibitors or antibodies can be used. Physical exercise is also an effective way of alleviating inflammatory cytokines, improving angiogenic functions and overall physical health [6].

Case presentation

In August 2016, a 26-year-old Mauritian female presented to a Rheumatologist for a private consultation in Port-Louis with symptoms of fatigue, unexplained weight loss, pain, weakness of the extremities, numbness, dizziness, a headache and arthritis. The provisional diagnosis was Rheumatoid Arthritis and she was prescribed corticosteroids as well as an immunosuppressant. However, the medications failed, and the patient also developed blurred vision, chest pain and an absence of her carotid pulse. She attended another doctor for a private consultation in July of 2017 and the diagnosis of TKA was made based on the clinical evaluation. An Immunology/serology (Table 1 and Table 2) test along with a duplex ultrasound scan was requested. (Figure 1-3).

As a follow up, both non-contrast and contrast enhanced CT scans were performed in 2019 and 2021 and revealed a peak flow velocity of the Right Carotid Artery to be 150cm/s, the Left Carotid Artery was 105cm/s and streamline flow was noted in Internal Carotid Artery and External Carotid Artery along with a reduced caliber. Focal thickening (3.6mm) with mural enhancement of the anterior wall of the descending aorta at the level of the crux of the diaphragm extending inferiorly over a length of 20 mm was noted. At the level of the renal artery, a focal thickening of 2.8mm extending inferiorly over 50mm was reported. She was also diagnosed with an autoimmune disease (Rheumatoid arthritis) in 2017. She has a negative medical history for diabetes, asthma, epilepsy, smoking, alcohol cerebrovascular apoplexy and any recreational drug abuse was ruled out. She was deficient in vitamin D. In March 2016 the patient had undergone a splenectomy. The patient had a drug history of Alendronate tablet (Bifosa) 70mg weekly, Chewable Calcium, Multivitamin tablets, Mycophenolic acid tablet (Cellcept) 500mg BD, Aspirin tab

75mg once daily, Atorvastatin tablet 20mg once daily and a Tenofovir tablet 300mg daily. There was no history of similar complaints in any of her family members.

Table 1: Serology/Immunology tests

Year	Anti Neutrophil Cytoplasmic Antibodies (ANCA)	Reference value
2017	C:4.20 P:0.35	Positive ≥ 1.10
2021	20	Negative < 20

Table 2: Changes in Diameter, intimal wall thickness, and peak flow velocity of vessels

	Year:2017	Year:2019	Year:2021
1. Right Common Carotid Artery			
-Diameter (6.0-8.0mm)	4.0mm	3.6mm	-
-Intimal wall thickening (less than 1mm)	1.1mm	2.0mm	-
Peak flow velocity (30-110cm/s)	150cm/s	-	-
2. Left Common Carotid Artery			
-Diameter (6.0-8.0mm)	3.8mm	2.7mm	3.0mm
-Intimal wall thickening (less than 1mm)	1.1mm	2.0mm	1.6mm
-Peak flow velocity (30-110cm/s)	105cm/s	-	-
3. Brachiocephalic trunk			
-Diameter (10-14mm)	4.5mm	-	-
-Intimal wall thickening	3.0mm	-	4.0mm
Less than 1mm			
4. Descending Aorta			
-at level of crus of diaphragm (less than 3mm)	-	3.6mm	-
-at level of origin or renal A (less than 2mm)	-	2.8mm	-
5. Subclavian Artery			
I) R. Subclavian Artery			
-Diameter (8-10mm)	-	4.5mm	-
-Thickness (less than 1mm)	-	2.0mm	-
II) L. Subclavian Artery			
Diameter (8-10mm)	-	-	-
-Thickness (less than 1mm)	-	-	1.9mm

Figure 1 (below) Depicts the Duplex ultrasound scan of the Right common carotid artery showing a reduced diameter of the lumen.

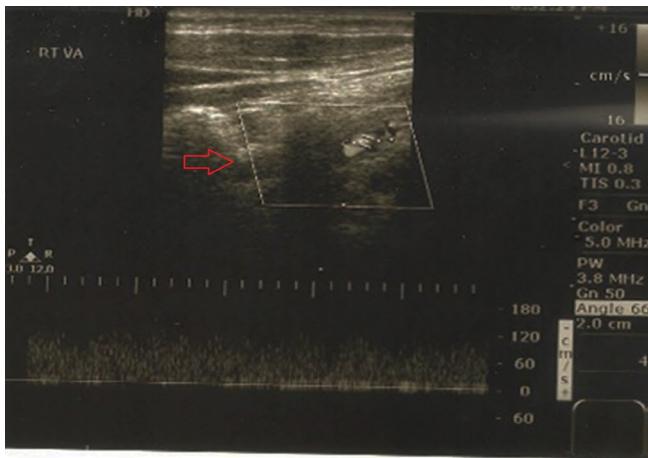


Figure 2 (below) Depicts the Duplex ultrasound scan of the right vertebral artery and the external carotid artery. Both showing a reduced lumen diameter.

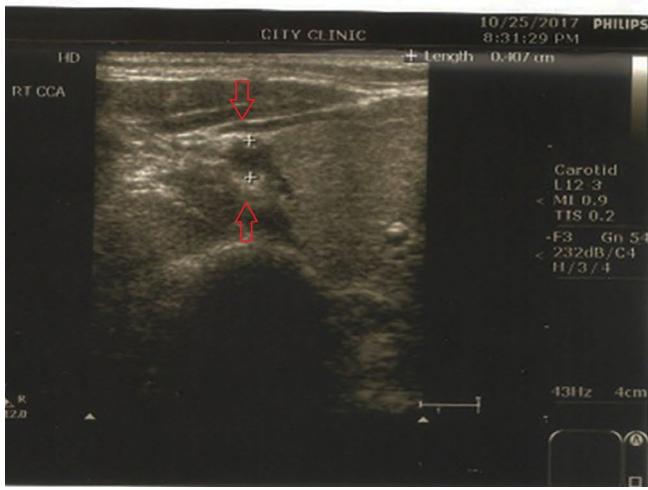
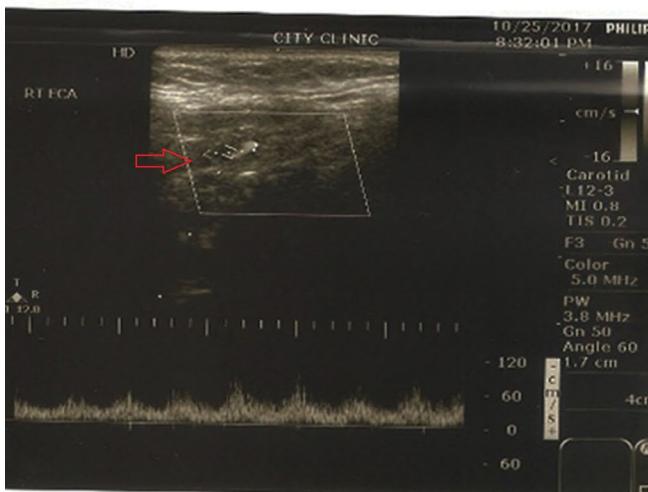
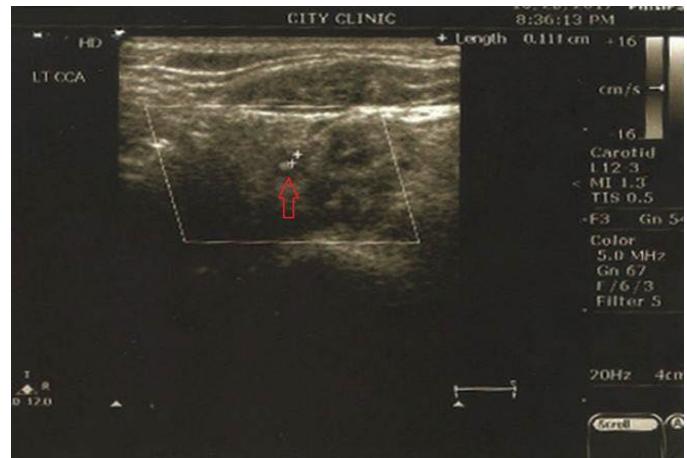
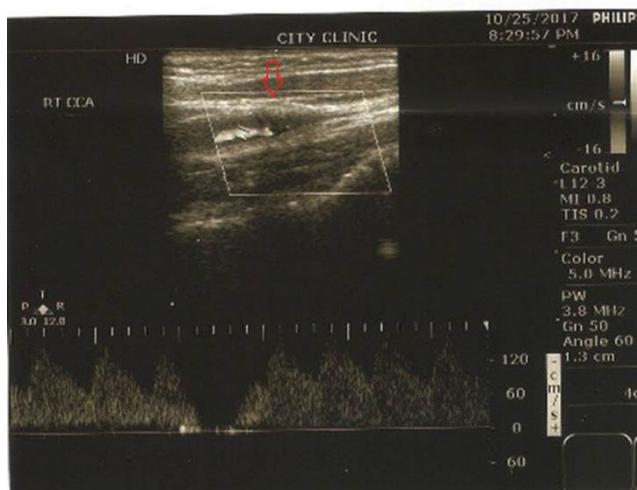
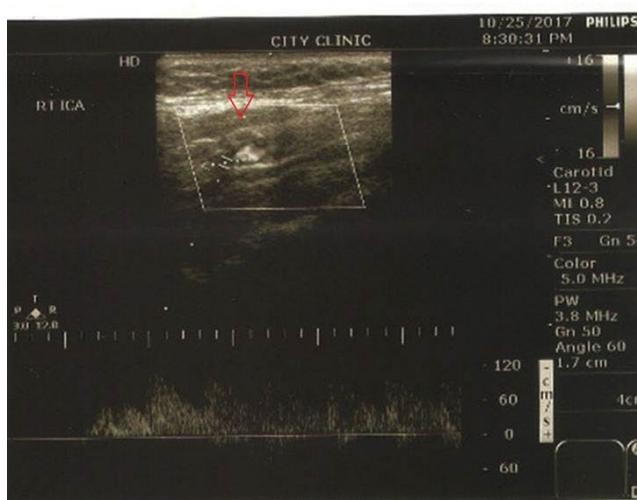


Figure 3 (below) Depicts the Duplex ultrasound scan of the left common carotid artery showing the “Macaroni” sign.



Discussion

According to the 2022 American college of Rheumatology (ACR) and the European Alliance of Associations for Rheumatology (EULAR) criteria, this patient meets the criteria for diagnosis. A score of ≥ 5 points is needed for the classification of Takayasu's arteritis [7]. Due to similar clinical features, TKA is often misdiagnosed as other types of vasculitis [7]. TKA can also be misdiagnosed as Giant Cell Arteritis (GCA) due to involvement of large vessels. Other differential diagnoses include Aortic Coarctation, Atherosclerosis, Behcet disease, Ig-G4 related diseases, Kawasaki disease, Rheumatoid arthritis or sarcoidosis [2]. As a general rule, the criteria are applied only if large vessel vasculitis (LVV) or medium vessel vasculitis is present. [1]. A detailed history, thorough clinical examination along with vascular physical examination and vascular imaging will aid in the diagnosis and differentiation between other vasculitis or GCA [7]. The diagnosis of TA is always a challenge. A case report by Yasuda T et al. showed a 50-year delay in the diagnosis of Takayasu Arteritis [8]. It is observed that by the time of diagnosis, the patient has already suffered significant morbidity with irreversible organ damage. In our case, the same scenario occurred where the patient's diagnosis was delayed and as a result, she suffered intimal thickening of various arteries. TKA is a chronic granulomatous inflammation, strongly associated with proinflammatory cytokines [4]. Hence, corticosteroids can be used. Nakaoka Y et al. has shown that a clinical response of 60% and return to normal of acute phase reactants occurred in 84% of patients [5]. Corticosteroids help to avoid vascular disease progression [4]. TNF- α inhibitors and IL-6 receptor antibodies are also used in combination with corticosteroids [3]. Surgery is also a modality of treatment. The surgical interventions depend heavily on where the lesions lie. For major aortic reconstruction, Dacron or Polytetrafluoroethylene are used. For isolated renal or mesenteric revascularization, autogenous transplant of the saphenous vein is used. In cerebrovascular insufficiency an aortic-carotid bypass is performed and for upper extremity ischemia, bypass distal to the subclavian, axillary or brachial arteries are performed. Furthermore, various aortic reconstructions are often performed. Surgery is normally well tolerated in young patients and dramatic postoperative improvements are observed [2]. Revascularization procedures can be performed for ischemic stroke, ischemic optic neuropathy and myocardial infarctions [5].

Clinical recommendation and long-term follow-up

Proper case histories can lead to an accurate diagnosis and should include the correct symptoms as well as duration, family history and thorough clinical examinations. Diagnosis should be made according to the 2022 criteria made by ACR and EULAR along with radiological imaging, computer tomography angiography (CTA), Magnetic Resonance angiography (MRA) and Colour

Doppler Ultrasound to assess the extent of the vascular involvement. MRA can be done with or without intravenous contrast as compared to CTA where intravenous contrast is required [5]. With imaging, the thickening of the walls of the blood vessels can be assessed. MRI, although recommended for TKA diagnosis, is costly and not readily accessible. Carotid artery ultrasound is thus commonly and easily performed as it is both low in cost, simple and noninvasive. Positive findings include a thickening of the wall of the Carotid artery. This is known as a Macaroni sign [9]. To evaluate the prognosis or post treatment efficacy, changes in CRP, ESR, IL-6 and TNF- α levels should be monitored [3]. In summary the most important factor for the early diagnosis of TA is the physician's awareness of the findings.

Nazareth and Mason proposed red flag signs which makes it easier to diagnosis and to identify the disease. These flags should be identified timely [5].

Table 3: Red flag findings for Takayasu arteritis in patients under the age of 40 years [10]

Carotidynia
Angina
Limb claudication
Absent/weak peripheral pulses
Hypertension
Discrepant blood pressure in the upper limbs (>10 mm Hg)
Arterial bruits
Aortic regurgitation
Unexplained acute-phase response (elevated erythrocyte sedimentation rate or C-reactive protein)

Conclusion

This report highlights the intricate process of diagnosing and managing TKA, accentuating the importance of thorough clinical assessments and meticulous history taking. By diving deep into the patient's medical background, signs and symptoms as well as with the proper investigations, healthcare providers can swiftly recognize the distinctive features of TKA. This report also highlights the long-term follow-up for disease progression and the efficacy of treatment for patients diagnosed with TKA.

Abbreviations

Takayasu's arteritis (TKA), Tumor Necrosis factor alpha (TNF- α), Interleukin (IL), Interferon gamma (IFN- γ), American college of Rheumatology (ACR), European Alliance of Associations for Rheumatology (EULAR), C Reactive protein (CRP), Right Subclavian Artery (R. Subclavian Artery), Left Subclavian Artery (L. Subclavian Artery)

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Authors' contribution

- study planning: KM, IB, MR, SR, JR, IB
- case report: KM, IB, MR, SR, JR, IB
- follow up: KM, IB, MR, SR, JR, IB
- interpretation: KM, IB, MR, SR, JR
- manuscript writing: KM, IB, MR, SR, JR
- manuscript revision: KM, IB, MR, SR, JR, IB
- final approval: KM, IB, MR, SR, JR, IB
- agreement to be accountable for all aspects of the work: KM, IB, MR, SR, JR, IB
- Supervisor: IB

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Availability of data and materials

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Consent for publication

All the authors confirm that patient's informed written consent was obtained for publication of clinical details and images of reports, with efforts are made to maintain the anonymity of the patient.

Competing interests

All authors declare no conflicts of interests or competing interests concerning the material in this manuscript.

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