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Case Report

Asymptomatic complete distal abdominal aortic occlusion with initial presentation of ruptured intracranial aneurysm *,**

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ABSTRACT

Aortoilliac occlusive disease is occlusive atherosclerosis disease involving the distal aorta and bifurcation of iliac arteries and it is a subtype of peripheral arterial disease. Total occlusion of the abdominal aorta is a rare occurrence with an incidence of 3% -8.5% among the aortoiliac occlusive disease patients. We present a case of a 53 years old patient with a background history of hypertension and ex intravenous drug abuser with negative retroviral screening status, with no previous complaints who was brought to the Emergency Department with sudden onset of altered sensorium and 1 episode of seizure. Computed tomography angiogram of the brain showed a ruptured anterior communicating artery aneurysm. Diagnostic conventional angiogram of the brain was planned; however, difficulty was encountered during bilateral femoral artery cannulation with the abrupt termination of bilateral common iliac arteries. Computed tomography angiogram of the abdomen showed incidental finding of total occlusion of the abdominal aorta. As a conclusion, total occlusion of abdominal aorta secondary to aortoiliac occlusive disease with an associated intracranial aneurysm is never reported in the literature to date. This case highlights the possibility of association in between these two conditions which may benefit from further research.

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Introduction

Peripheral artery disease (PAD) is a global issue with global prevalence between 3 to 12 percent [1]. Aortoilliac occlusive disease(AOD) is occlusive atherosclerosis disease involving the distal aorta and bifurcation of iliac arteries and it is a sub-type of PAD [2]. Total occlusion of the abdominal aorta(TOAA) is a rare occurrence with an incidence of 3% -8.5% among the aortoiliac occlusive disease patients [3]. There is no reported association between peripheral arterial disease and intracranial aneurysm. We present a rare case of a 53 years old patient who initially presented with ruptured anterior communicating artery aneurysm with incidental finding of total occlusion of the abdominal aorta. We discuss the pathophysiology, imaging and management of these rare conditions.

Case description

A 53-year-old male with a background history of hypertension, non-compliant to medication was presented to a district hospital with a history of sudden onset of altered sensorium and 1 episode of seizure. He had severe headache episode before the start of the symptoms. The patient is also an ex-intravenous drug abuser with negative retroviral screening status.

On initial physical examination, the patient's conscious level was recorded as E1V3M5 based on Glasgow's coma scale (GCS) with a blood pressure of 230/150 mmHg and a heart rate of 58 beats per minute (bpm). Central nervous examination reveals hypotonia and hemiparesis of the left side of his body with the power of 0/5. The tone was normal on the right side, with power slightly reduced measuring 3/5. Twelve-lead ECG shows normal sinus rhythm and heartbeat of 65 bpm. Oxygen saturation was maintained at 100% via a high flow mask. Apart from the high urea and creatinine levels, the rest of his initial haematological and biochemical parameters were normal. He was electively intubated for cerebral protection due to a significant drop in GCS to 5/15 after admission. Based on the history and clinical examination, a primary diagnosis of cerebral stroke was made, and the patient was subjected to urgent computed tomography (CT) scan of the brain. It showed massive right frontal intraparenchymal haematoma (Fig. 1a) and diffuse subarachnoid haematoma (Fig. 1b), which was complicated with local mass effect and generalized oedema. He was then referred to our hospital for further management by our neurosurgical team.

At our centre, an urgent computed tomography angiogram (CTA) of the brain was done, which reveals a ruptured saccular aneurysm of the anterior communicating artery with a narrow neck (Fig. 2). Due to extensive intraparenchymal haematoma, an urgent diagnostic subtraction angiogram of the brain (DSA brain) was planned to map the aneurysm and look for other cerebral vascular abnormalities.

However, there was a difficulty of cannulation of the abdominal aorta from the bilateral femoral arteries access site. A bilateral ascending femoral angiogram via the access site reveals abrupt termination of the bilateral common iliac arteries with multiple collateral vessels arising from these iliac

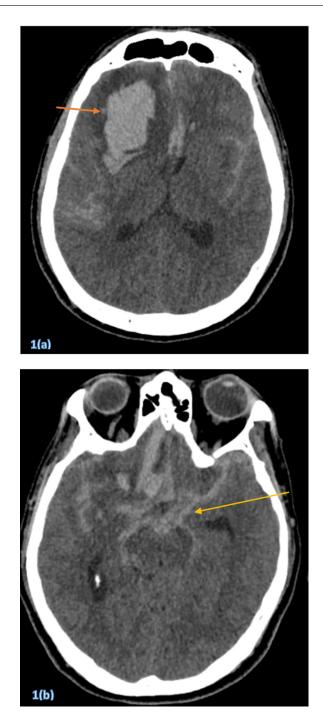


Fig. 1 – Non-contrast Computed Tomography (CT) of brain showing (a) right intraparenchymal haemorrhage in the right frontal lobe (Orange arrow) with generalized cerebral oedema and (b) diffuse subarachnoid haemorrhage occupying the interpeduncular cistern (Yellow arrow) and bilateral Sylvian fissure. (Color version of the figure is available online)

arteries (Fig. 3). Thus, a preliminary diagnosis of stenosis or thrombosis of the bilateral iliac arteries was made. A CTA of the abdominal and pelvis shows a calcified stenosed abdominal aorta at the level of L3 (Fig. 4) with the intraluminal thrombosis (Fig. 5a and 5b) causing total occlusion of the lumen. The thrombus extends up to the level of L1, stopping just inferior

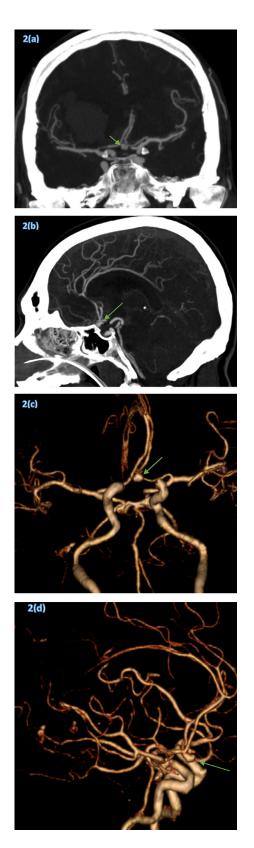


Fig. 2 – Computed Tomography Angiogram (CTA) of the brain in coronal view(a), coronal view (b), and 3D reconstruction [(c)&(d)] showing the saccular aneurysm of the anterior communicating artery.



Fig. 3 – Digital subtracted angiogram (DSA) from the (a) right and (b) left femoral arteries show abrupt tapering of the bilateral common iliac arteries with no further contrast opacification beyond the obstruction site (yellow arrow). There were multiple collateral vessels noted bilaterally. (Color version of the figure is available online)

to the origin of the superior mesenteric artery (SMA). Celiac, SMA and hepatic arteries are of normal calibre. However, the inferior mesenteric artery is thrombosed. Tight stenosis of the proximal right renal artery with the narrowest diameter measuring 0.1cm. However, prompt perfusion of the right renal. A small left renal artery arises from the SMA supplying a hy-

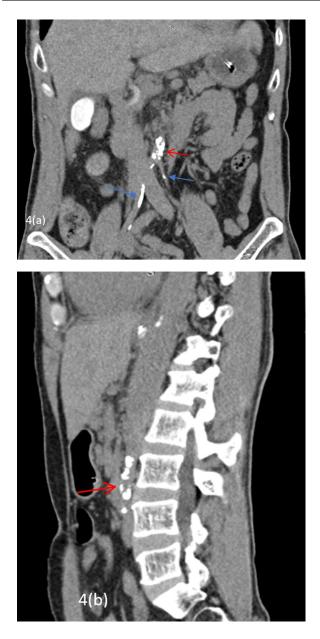


Fig 4 – (a &b). Non-contrast CTA of the abdominal aorta in coronal(a) and sagittal images (b) showing heavily calcified vessel within the distal abdominal aorta at the L3 level (red arrow). Scattered wall calcification of the bilateral common iliac arteries (blue arrow). (Color version of the figure is available online)

poplastic left kidney (Fig. 5d). Bilateral femoral are retrogradely opacified from the tortuous collaterals arising from SMA via mesenteric vessels suggesting visceral collaterals. The visualized descending thoracic aorta was normal in calibre.

A CTA of the thoracic aorta was planned once the patient's condition was stable to assess the rest of the aorta thoroughly. Craniotomy, clot evacuation and clipping of an aneurysm were also planned, but the family members were not keen on surgical intervention. Unfortunately, the patient's condition deteriorated, and he succumbed to complications of intracranial haemorrhage. Fig. 5 – (a-d). Post-contrast CTA of the abdominal aorta in coronal(a) and sagittal images (b) showing complete obstruction of the distal abdominal aorta (red arrows) below the SMA proximal to the heavily calcified distal abdominal aorta (blue arrow). Coronal image (c) showing large tortuous inferior epigastric arteries (orange arrow) are supplying both iliac arteries. Coronal image (d) shows an incidental finding of a small-sized left kidney (yellow arrow) with a normal-sized right kidney (green arrow). (Color version of the figure is available online)

Discussion

Peripheral artery disease (PAD) is a global issue with global prevalence between 3 to 12 percent. Prevalence of patients with PAD predominantly from low/middle income regions, with 55 million patients coming from southeast Asia. Another issue is the rise in the number of patients affected by PAD which increased by 29 percent in low/middle income regions in a duration of a decade, from the year 2000 to the year 2010, when compared with the preceding decade [1].

Depending on the location of the occlusion, PAD is broadly divided into femoropopliteal, aortoiliac and tibiofibular disease [4,5]. Aortoiliac occlusive disease(AOD) is occlusive atherosclerosis disease which causes progressive narrowing of the distal abdominal aorta, iliac and femoropopliteal arteries [2]. Symptomatic AOD usually presents a classical triad of bilateral lower limb claudication, absent or decreased femoral pulsation and erectile dysfunction [5]. Surprisingly, many patients remain asymptomatic because of extensive collateral vessels [6], as seen in our case.

Total occlusion of the abdominal aorta (TOAA) is a rare disease. Infrarenal TOAA has an estimated incidence of 0.15% in the autopsy study [7], and the incidence rises to 3% -8.5% among the aortoiliac occlusive disease patients [3]. Rene Lireche reported the first chronic infrarenal aortic occlusion in 1923 [8]. TOAA could be either an acute or a chronic onset disease. Acute onset disease tends to have a catastrophic outcome, including death in some cases. The main cause of acute obstruction is embolic occlusion or thrombotic events, mainly due to severe atherosclerosis [9]. Meanwhile, chronic occlusions are more tolerated as the body adapts to the changes in the vascular flow by developing collateral blood supply which maintains the basal perfusion [10]. Chronic occlusion of the aorta secondary to atherosclerosis tends to occur in the late advanced stage [10]. Fifty percent of the chronic TOAA presents as juxtarenal or para-renal occlusion [3].

Conventional aortography is the gold standard for evaluating aortic stenosis [11]. However, the presence of distal TOAA could cause a problem in cannulation and complete visualization of the occlusion if the femoral access is used, as in our patient. In such cases, angiography via radial or brachial arteries would be an answer, more so in our patient, as there is a need to image the cerebral vessels.

Another modality, multisection CT, is currently gaining momentum in the diagnosis of aortic stenosis and occlusive aortic diseases compared to conventional aortography. It allows visualization of the aorta, and its branches and collaterals as well as wall calcification and intraluminal thrombus in a single session. CT is also useful for looking at the causes, examine the para-aortic region and follow up patient postintervention [11,12]. The imaging is even faster and provides high-quality arterial phase data which could be used for threedimensional reformations. Such reformations are very useful in pre-operative planning [11]. Other useful modality includes Doppler ultrasonography. It is good for the assessment of vessel wall thickness and signs of vessel inflammation, like the one caused by vasculitis [12].

Treatment of TOAA includes medical management of the symptoms, especially hypertension and vasculitis [13]. Primary definitive management of aortic occlusion is the surgical intervention which includes aorta-aortic bypass, patch aortoplasty, aortic endarterectomy, aorto-bi femoral bypass, extra-anatomical bypass and visceral artery reconstruction [3,8,9,13]. The success rate of surgery is good, with normalization of the blood pressure is noted in up to 70% of cases. However, in the case of infective or inflammatory cause of stenosis, the surgery is recommended to be postponed until the active phase of vasculitis is over[14]. Surgical management of the chronic infrarenal and suprarenal chronic aortic occlusion have peri-procedural mortality of approximately 5% and 23% respectively [9,15]. Surgical treatment is associated with longer hospitalization and longer recovery[7]. Meanwhile, endovascular intervention is also gaining some momentum in providing minimally invasive intervention in a selected group of patients especially those without mesenteric or renal arteries involvement [8,13]. The technical success of endovascular intervention of infrarenal TOAA is 73% - 93% [7]. Severe aortic calcification is the independent determinant of interventional failure [7]. Hemodynamic of the aorta and cardiovascular system shows improvement immediately after revascularization in the case of infrarenal TOAA [3]. Therefore, with proper management, the clinical condition of the patient could be improved, and severe complication prevented.

There is no established correlation between peripheral arterial disease with intracranial aneurysm in published literature. The intracranial saccular aneurysm has an incidence of 1% -2% in general populations [16]. Unruptured cerebral aneurysms are found in 3 – 8% of the general population [17]. The Anterior Communication artery (ACom) is one of the common sites for the cerebral aneurysm, especially in adults [16,18]. ACOM aneurysm is also associated with a higher risk of rupture [17]. Subarachnoid haemorrhage (SAH) secondary to aneurysm rupture associated with high morbidity and mortality. ACom aneurysm is diagnosed in 22% -30% of patients presented with SAH [17].

In conclusion, TOAA secondary to aortoiliac occlusive disease with an associated intracranial aneurysm is never reported in the literature to date. This case highlights the possibility of association between these two conditions which may benefit from further research.

Author contribution

CN and AAW prepared and wrote this article. CN and JH involved in managing the patient, preparing the figures, providing the radiological descriptions and editing the final manuscript. CYN wrote and revised the manuscript as well as acted as the corresponding author. All authors have access to the manuscript and had agreed to publish it as in the current form.

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Patient consent

Written informed consent was obtained from the patient's next of kin for the publication of this case report.

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