Widespread involvement of hepatic, renal and mesenteric arteries with multiple mycotic aneurysms in a child

Sibel Kul[1], Aydın Aydın[2], Hasan Dinç[1], Erol Erduran[3]

Departments of [1]Radiology and [3]Pediatrics, Karadeniz Technical University, Faculty of Medicine, and [2]Department of Urology, State Hospital, Trabzon, Turkey

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Multiple visceral microaneurysms of mycotic origin are very uncommon. We present an 11-year-old child with the clinical and biochemical signs of septicemia in whom arteriographic study revealed multiple microaneurysms of renal, hepatic, gastroduodenal, ileocolic and right colic arteries. Six weeks of antibacterial treatment resulted in resolution of the septicemia and most of the aneurysms healed without the need for endovascular or surgical treatment.

Key words: mycotic aneurysm, visceral arteries, mesenteric arteries.

Aneurysms are rarely seen in children. Multiple visceral microaneurysms is a characteristic finding of polyarteritis nodosa, but have also been reported in other systemic diseases, such as systemic lupus erythematosus, Wegener's granulomatosis, fibromuscular hyperplasia and Henoch-Schönlein purpura, and after viral infections or trauma[1,2]. Infections causing septic embolism can also result in aneurysm formation named as mycotic aneurysm.

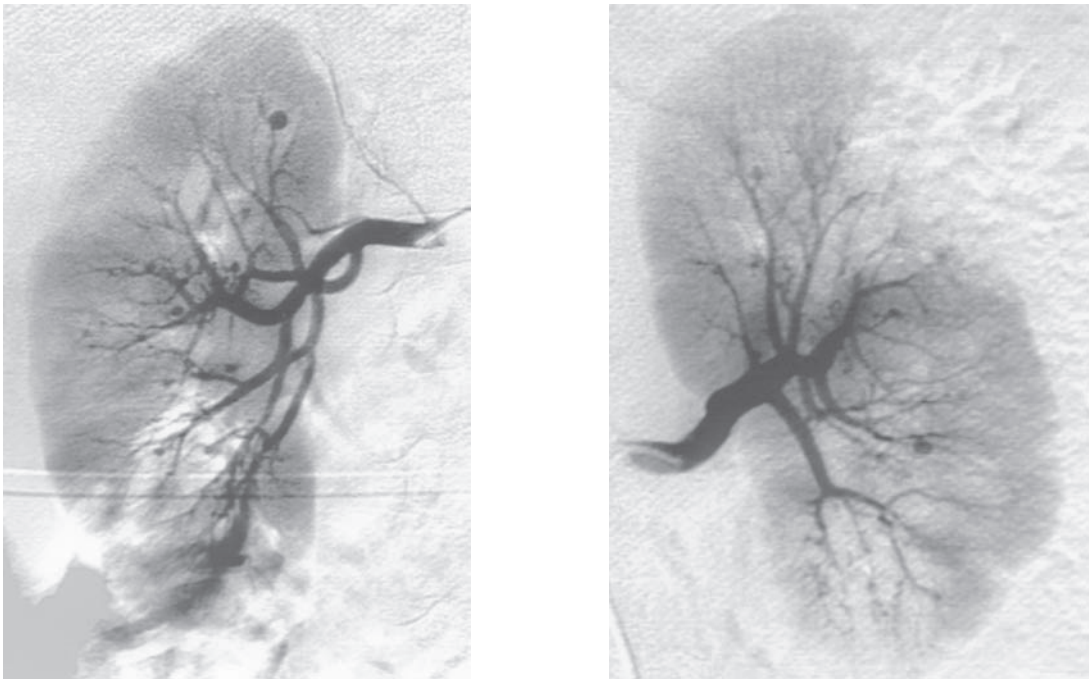
Mycotic aneurysms are life-threatening because of their high potential to rupture and to cause organ or limb ischemia. Thus, early treatment is necessary. Anti-biotherapy, endovascular treatment and surgery are the possible treatment options; however, even with treatment, their evolution is uncertain and associated with high mortality.

To the best of our knowledge, such widespread involvement of visceral arteries with multiple mycotic microaneurysms has not been reported before.

Case Report

An 11-year-old boy was admitted to our hospital with fever and generalized convulsion. At admission his fever was 39.5ºC with normal neurological findings.

Laboratory testes revealed anemia with hemoglobin level of 9.8 g/dl and leukocytosis with white cell count of 12,000 g/dl. Erythrocyte sedimentation rate and C-reactive protein level were increased. Urine analysis revealed microscopic hematuria. Liver function tests were abnormal with increased alanine and aspartate aminotransferase levels. Azotemia was diagnosed with high serum urea and creatinine levels. His blood pressure was high (130-95 mmHg). Echocardiography revealed left ventricular dilatation, mitral valve vegetations, mitral and tricuspid regurgitations and thrombus. Blood culture obtained at admission grew Staphylococcus aureus. Cranial magnetic resonance imaging (MRI) scans were normal. Convulsion did not repeat again. The patient received antihypertensive (captopril, nifedipine), anticoagulant (warfarin sodium) and antibacterial (vancomycin, amikacin) treatments. One week later the patient developed macroscopic hematuria. With the suspicion of vasculitis, first the renal - because the kidney is the most commonly affected organ – and after celiac, mesenteric, and cerebral arteriographies were performed via transfemoral accesses. Figures 1A and B are selective renal intra-arterial digital subtraction angiographies (DSA) showing multiple small-



(a) (b)

Fig. 1. a, b. Bilateral selective renal arteriograms show diffuse multiple intraparenchymatous saccular aneurysms and wedge-shaped hypoperfused areas in both kidneys.

sized saccular aneurysms in the distal branches of bilateral renal arteries. Selective celiac arteriogram (Fig. 2) revealed multiple saccular microaneurysms in the distal branches of the right and left hepatic arteries and one in the proximal gastroduodenal artery. Selective superior mesenteric arteriogram (Fig. 3) revealed a few similar microaneurysms in the distal right colic and ileocolic arteries. Although rupture of one of the renal aneurysms in to the collecting system was thought to be the cause of hematuria, no contrast extravasations

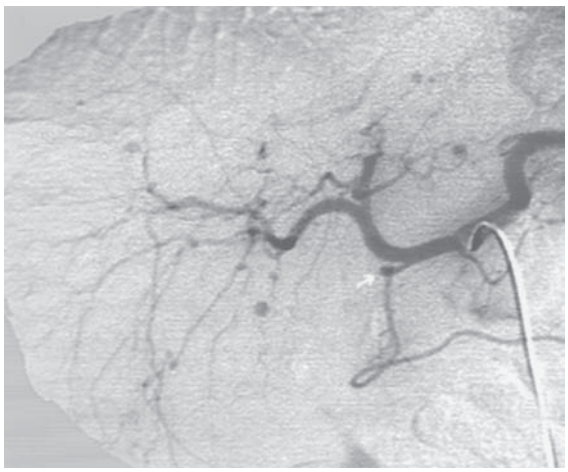


Fig. 2. Celiac arteriogram reveals multiple saccular microaneurysms in the distal branches of right and left hepatic arteries and one in the proximal gastroduodenal artery (arrow).

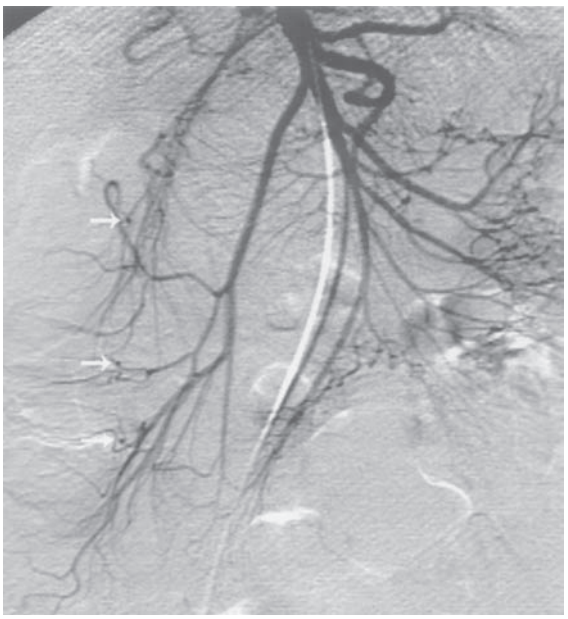


Fig. 3. Superior mesenteric arteriogram shows microaneurysms (arrows) in the distal right colic and ileocolic arteries.

from the renal aneurysms were detected. There was no aneurysm in splenic, other mesenteric or cerebral arteries. There were no signs or symptoms of polyarteritis nodosa. The results of serological panel for a collagen vascular disease or an autoimmune disorder (antinuclear antibodies, antineutrophil cytoplasmic antibody) were negative. Serological tests for hepatitis were normal. With clinical, biochemical, blood culture and echocardiographic findings, these multiple aneurysms were thought to be infectious in origin.

After the six-week antibiotic treatment, the patient remained afebrile, hematuria resolved and renal and hepatic functions and hypertension improved. A second arteriogram performed 10 weeks later showed only a few residual microaneurysms in both renal intraparenchymal arteries (Figs. 4A, B) and right segmental hepatic arteries (Fig. 5). The patient is under clinical follow-up for these remaining aneurysms.

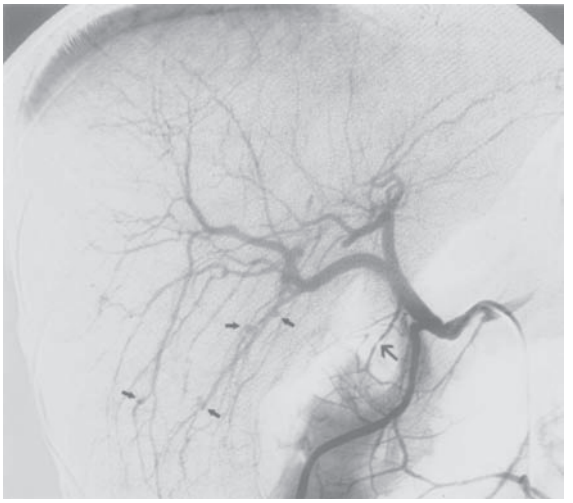
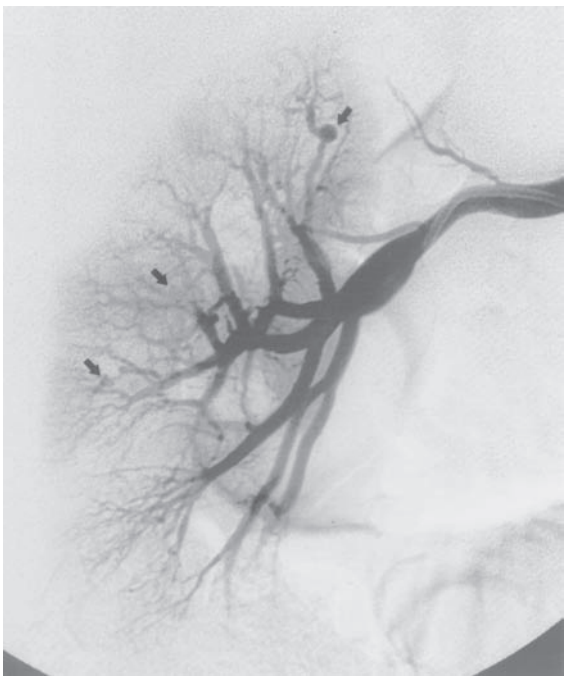
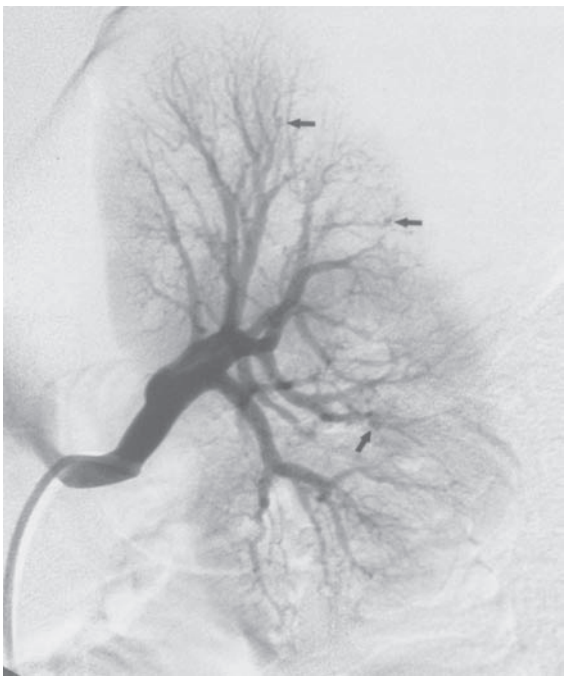


Fig. 5. Control selective hepatic arteriogram shows complete loss of the proximal gastroduodenal artery aneurysm with formation of a short segment focal stricture (large arrow). Most of the hepatic artery aneurysms had resolved, with only a few residual ones (small arrows) in the right hepatic artery.



(a)



(b)

Fig. 4. a, b. Control selective renal arteriograms obtained 10 weeks after initial arteriograms show few residual intraparenchymal aneurysms (arrows) in both kidneys.

Discussion

Aneurysms seen in children should be carefully evaluated for an underlying secondary disease, history of trauma or septicemia[3]. They all produce similar angiographic appearance of aneurysmal dilatation and it can be difficult to differentiate the underlying cause. In this case, the child had no history of clinically important abdominal trauma. The hepatitis serologic tests and autoimmune serologic panel were

normal. Presence of febrile illness, findings of endocarditis and positive blood culture made infection the likely cause for multiple aneurysms. Distal location of aneurysm, presence of arterial occlusion or stenosis close to the aneurysm, multiple aneurysms and rapid morphologic changes also help in the diagnosis of infective etiology. Infected aneurysm of any cause is termed as mycotic aneurysm. Acute bacterial endocarditis is the most common cause of mycotic aneurysms and mycotic aneurysms develop in 3-15% of patients. Septic embolization to the vasa vasorum of arteries cause vessel wall necrosis and aneurysm formation[4,5].

Mycotic aneurysms usually occur in the aorta and, as primary branches of the aorta, the femoral, cerebral and visceral arteries are the next most common sites[3]. Most reported cases of visceral mycotic aneurysms are in the form of a single or a few aneurysms involving one of the visceral territories. Appearance of multiple visceral mycotic aneurysms in the same patient as seen in polyarteritis is extremely rare. Such cases are reported by Mourad et al.[6] as multiple renal microaneurysms related to bacterial endocarditis and by Tihansky et al.[7] as multiple mycotic splenic artery aneurysms. Also, Charlier et al.[8] reported one hepatic aneurysm with multiple renal aneurysms of mycotic origin. Our case is unique because of such extended involvement of hepatic, renal and mesenteric territories.

Anti-biotherapy, transcatheter embolization and surgical resection with vascular reconstruction are the possible treatment approaches for mycotic aneurysms. The most common approach for visceral and intracranial mycotic aneurysms in recently reported cases is antibacterial treatment of underlying infection and then percutaneous embolization or surgery[4,9,10]. Antibiotic treatment may reduce the hemorrhagic risk related to mycotic aneurysms and even some mycotic aneurysms may resolve. But it is impossible to predict the outcome. Even in the same patient with multiple mycotic aneurysms, the response of each aneurysm to anti-biotherapy may be different. In an extensive review of the literature, during or after the anti-biotherapy of the mycotic aneurysms, complete regression was found in 20, rupture in 17, enlargement of aneurysm in five and de nova aneurysm formation in five reports[9]. There are no certain predictive factors to evaluate the risk of rupture. In mycotic aneurysms presented with rupture, emergent treatment with transcatheter embolization without early use of antibiotic therapy is required[4]. However, whether or not to use anti-biotherapy or endovascular treatment for those unruptured mycotic aneurysms remains controversial. Although transcatheter embolization is a minimally invasive and effective treatment approach in experienced hands, risk of residual infection of the aneurysm after the insertion of embolization materials is a frightening complication[4]. There are only single cases of reported peripheral mycotic aneurysms successfully treated with embolization either by coils or particles[11-14]. Surgical resection of the aneurysms with vascular reconstruction is a commonly used approach in mycotic aneurysms of large arteries and is not suitable for multiple intraparenchymatous microaneurysms with multiorgan involvement. Because of the multiplicity of small-sized aneurysms and no appearance of active bleeding in angiography, we did not perform embolotherapy in our patient and only used antibacterial treatment. This treatment regimen provided complete resolution of most of the aneurysms. Although selective transcatheter embolization might be a choice of treatment for the remaining aneurysms after the completion of anti-biotherapy, we chose clinical follow-up. The patient has been symptom-free for more than a year.

In conclusion, although multiple microaneurysms involving visceral organs and gastrointestinal tract are a well-known finding during the course of polyarteritis, they also rarely can be of infectious origin. Because their treatment approaches are very different, the two must be differentiated, and for such multiple small-sized mycotic aneurysms, antibacterial treatment must be the first treatment option.

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