

CASE REPORT

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Superior mesentery artery aneurysm with thrombosis and proximal stenosis in a case with Ehlers–Danlos syndrome

Mehmet Eren Ozturk, Ahmet Yalcin

ABSTRACT

Introduction: Visceral arterial aneurysms are very rare in the pediatric patient population and might be associated with connective tissue disorders.

Case Report: In this report, we present a 14-year-old male case with a superior mesenteric artery aneurysm with Ehlers–Danlos syndrome detected with computed tomography (CT) angiography. The patient had a history of scoliosis surgery associated with Ehlers–Danlos syndrome. A superior mesenteric artery aneurysm was detected along with a distal partial thrombosis and proximal stenosis. The patient was deceased 24 hours after admission to emergency room (ER) due to a generalized intestinal infarct.

Conclusion: Visceral arterial aneurysms are very rare and associated complications might be fatal in the child population. Computed tomography (CT) angiography imaging provides a fast and accurate diagnosis and can delineate additional prognostic findings.

Keywords: Computed tomography, Ehlers–Danlos, Intestinal ischemia, Superior mesenteric artery aneurysm Stenosis, Thrombosis

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INTRODUCTION

Arterial aneurysms of the major arteries are common in the adult population. However, aneurysms involving the visceral arteries are rare with an incidence of 0.01% [1, 2]. Superior mesenteric artery (SMA) related aneurysms constitute a small portion of the visceral aneurysms and have a mortality rate up to 35% [3]. Visceral arterial aneurysms are very rare in the pediatric patient population and might be associated with connective tissue disorders [4]. In this report, we present a peculiar case with SMA aneurysm, distal partial thrombosis and proximal stenosis in a 14 years old male detected by computed tomography angiography (CTA).

CASE REPORT

A 14-year-old patient presented to the emergency department complaining of sudden onset of cramping abdominal pain around the navel and deterioration in general condition 6 hours prior. He had a previous scoliosis operation at the age of 10 and diagnosis of Ehlers–Danlos type IV disease. On initial examination, the skin was moist and pale, blood pressure was hypotensive at 85/45 mmHg, pulse was 125 indicating tachycardia, and respiratory rate was 24 indicating tachypnea. Abdominal examination showed tenderness and rebound. Abdominal ultrasound showed collection of fluid in the peritoneal cavity and an abnormal dilated vessel anterior to the aorta. Doppler examination showed vascular color-coding consistent with vascular filling in the abnormal dilated vessel. The patient was referred to the CT unit for the detailed evaluation of abdominal circulation by contrast-enhanced CTA. On CT images, a proximal stenotic segment and aneurysm of the SMA was detected (Figure 1A and B). A partial thrombus was detected distal to the dilated segment (Figure 2A). More caudal images showed intraperitoneal fluid and

hypoperfusion of ileal and jejunal loops (Figure 2B). The patient underwent laparotomy in which subtotal intestinal ischemia was detected. The patient deceased in post-operative intensive care unit due to generalized intestinal infarct at 24 hours after admission to ER.

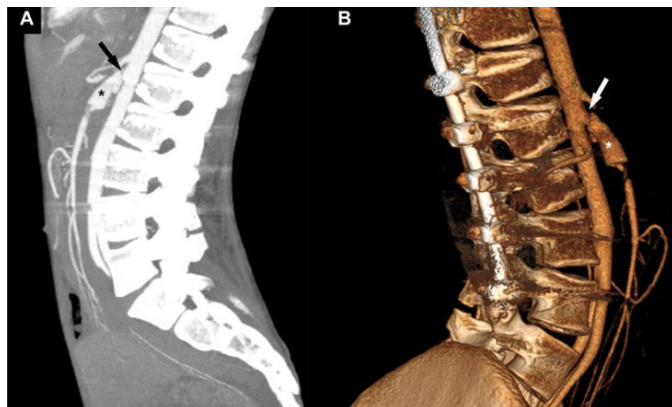


Figure 1: Sagittal MIP CT image (A) and 3D volume-rendered image (B) show the aneurysm of the SMA (asterisks) and proximal stenosis (arrows).

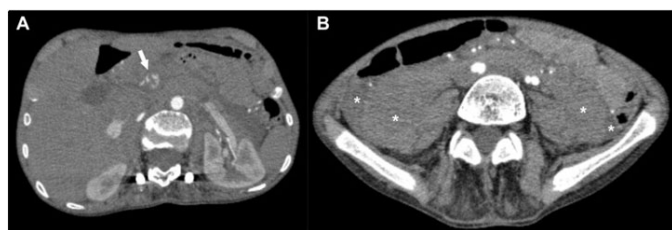


Figure 2: Axial contrast-enhanced CT image (A) acquired from the lower level of the aneurysm shows thrombosis obscuring the luminal filling of the SMA (arrow). More caudal image (B) shows hypoperfused intestinal loops and peri-intestinal fluid (asterisk).

DISCUSSION

Our case had proximally orientated SMA aneurysm and the distal thrombus which were concluded with fatal intestinal ischemia. Our case also had a stenotic segment proximal to the aneurysm. The formation of visceral aneurysms distal to the stenotic segment has been associated with high and low wall shear stress theories. These theories may explain the pathophysiology of the visceral aneurysm that formed distal to the stenotic segment in our case [5].

Pediatric non-aortic arterial aneurysms are extremely uncommon. Several factors such as infection, trauma, connective tissue diseases, noninfectious arteritis, or congenital vascular malformations are responsible for the development of aneurysm in the child population [6]. Aneurysms involving the visceral arteries are extremely rare in the pediatric population. These aneurysms can be due to fungal or bacterial infection, noninfectious inflammatory disease, or connective tissue disorders [7, 8]. Except for connective tissue related syndromes such

as Ehlers–Danlos type IV disease, surgical repair is the main treatment option. However, in cases with Ehlers–Danlos syndromes, surgery is not preferable due to fragility of the vessels and ligation of the dilated vessel is advised [7, 9].

Our patient had a history of scoliosis operation and diagnosed with Ehlers–Danlos syndrome. Furthermore, the patient had a pre-aneurysmal stenotic segment and thrombosis in the distal segment of the aneurysm. Detection of thrombosis was important as it determined the fate of our patient. Thrombosis of the celiac axis was reported in another case who had type IV Ehlers–Danlos and underwent an operation of correction of scoliosis. In that case, authors associated the thrombosis with surgery [10].

Contrast-enhanced CT angiography was extremely useful in this case, as it showed both the aneurysm and distal thrombus which allowed the surgeons to take necessary steps after definitive diagnosis. Doppler ultrasound imaging was used as a first step imaging method without radiation exposure. The diagnosis was made quickly and accurately in the emergency room, but the patient's delay in presenting for treatment 6 hours later delayed the early diagnosis and revascularization that are crucial in mesenteric ischemia, leading to the patient's death due to intestinal necrosis [11].

CONCLUSION

Visceral arterial aneurysms are very rare and associated complications might be fatal in the child population. Computed tomography angiography imaging provides a fast and accurate diagnosis and can delineate additional prognostic findings.

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Author Contributions

Mehmet Eren Ozturk – Conception of the work, Design of the work, Acquisition of data, Interpretation of data, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Ahmet Yalcin – Conception of the work, Design of the work, Analysis of data, Drafting the work, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Guarantor of Submission

The corresponding author is the guarantor of submission.

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Conflict of Interest

Authors declare no conflict of interest.

Data Availability

All relevant data are within the paper and its Supporting Information files.

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