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CASE REPORT

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Percutaneous Transcatheter Embolization of Gastrointestinal Bleeding in a Child with Polyarteritis Nodosa

Authors' Contribution:
A Study Design
B Data Collection
C Statistical Analysis
D Data Interpretation
E Manuscript Preparation
F Literature Search
G Funds Collection

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Summary

Background: Polyarteritis nodosa is a form of necrotizing vasculitis of small and medium-sized arteries. Major gastrointestinal complications are ulceration, perforation, hemorrhage, and obstruction.

Case Report: We report on a clinical case of a 16-year-old female patient with massive hematemesis, who was successfully treated with embolization with a 1:2 dilution of N-butyl cyanoacrylate glue.

Conclusions: To the best of our knowledge, this is the youngest child reported on with massive GI bleeding secondary to PAN, treated with successful percutaneous transcatheter embolization under emergency conditions.

MeSH Keywords: Anti-Neutrophil Cytoplasmic Antibody-Associated Vasculitis • Embolization, Therapeutic • Polyarteritis Nodosa

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Background

Polyarteritis nodosa (PAN) is a necrotizing angiitis that predominantly affects small- and medium-sized arteries [1,2]. PAN occurs rarely during childhood. Boys and girls seem to be equally affected, with a peak age of 10 years [3]. Multiple organ systems, such as renal, musculoskeletal, gastrointestinal, cardiovascular, and central nervous systems, and skin may be involved. Etiology is unknown, and ensuing vasculitis may result in thrombosis or aneurysm formation in any organ of the body [4]. The diagnosis is ideally established on the basis of a biopsy of involved tissue in a patient with appropriate clinical symptoms and laboratory data, but an angiogram provides a proof in some cases [5]. The prognosis of untreated PAN is very poor [6] and it depends on the presence and severity of visceral involvement. Diagnosis of PAN is sometimes difficult because symptoms are diverse and no specific serological test exists so the disease may not be recognized until the last stage. We present a case of massive gastrointestinal (GI) bleeding associated with PAN diagnosed and treated with endovascular approach.

Case Report

A 16-year-old girl presented with massive hematemesis to the emergency department of our hospital. The patient had pallor, tachycardia (120 beats/minute), tachypnea (25 respirations/minute), and hypotension (blood pressure, 70/40 mmHg). Laboratory investigation showed ahemoglobin level of 5.1 g/dL. Other laboratory parameters were normal. During the workup period, she received transfusion of 2 units of packed red blood cells, but control of bleeding could not be achieved. Findings of emergency upper GI endoscopy were completely normal, except for luminal blood. The 3D-CT angiography, a noninvasive imaging technique, is helpful in assessing the distribution of vessel involvement in PAN [7] but because of massive blood loss, emergency angiography was planned for detection of the bleeding site and possible endovascular treatment. Abdominal aorta angiography was performed with a 5-F pigtail catheter after a right common femoral artery puncture. One microaneurysm was seen in the left hepatic artery and another one in the intraparenchymal segments of the right renal artery (Figure 1A). Selective left gastric artery injection with a 4 French USL catheter revealed active bleeding in the fundus of the stomach. A

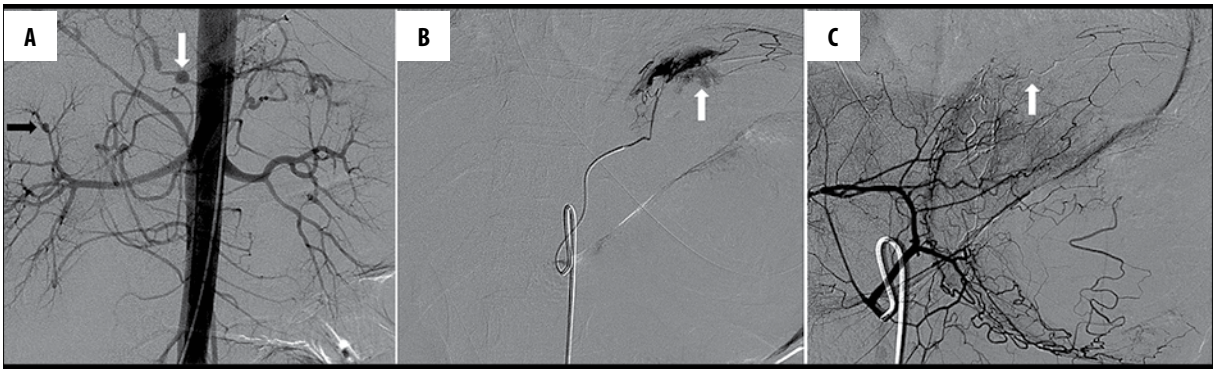

Figure 1. (**A**) An Injection to the abdominal aorta shows a microaneurysm of the left hepatic artery (white arrow) and intraparenchymal segment aneurysm of the right renal artery (black arrow). (**B**) Selective injection with a microcatheter shows contrast agent extravasation distal to the left gastric artery (white arrow). (**C**) An injection to the left gastric artery after embolization shows an active bleeding vessel occluded without contrast agent extravasation (white arrow).

2.7-F microcatheter was used in a coaxial fashion to select a vessel with contrast agent extravasation (Figure 1B). The bleeding vessel was occluded by injection of 1.5 mL of N-butyl cyanoacrylate glue diluted 1:1 with Lipiodol. Before injection of the glue mixture, the lumen of the microcatheter was flushed with 5% dextrose solution. Postembolization angiography demonstrated total occlusion of the active bleeding vessel (Figure 1C). There were no periprocedural complications and no postprocedural clinical evidence of ischemia. After the procedure, the patient's hemodynamic parameters stabilized.

Discussion

Gastrointestinal tract involvement is seen in 23% to 80% of patients with PAN and the major gastrointestinal complications are ulceration, perforation, hemorrhage, and obstruction [4]. Catastrophic and massive intestinal bleeding is rare unless an aneurysm ruptures in the intestine. The mortality rate can be as high as 75% in these cases [3]. Prognosis of PAN depends on the presence and severity of visceral involvement. The presence of multiple microaneurysms in the liver, kidney, and other viscera is common. Although some progress has been seen in the diagnostics and treatment of PAN in the last 2 decades, several studies from that period reported extremely high mortality rates in patients who developed acute abdomen [4]. Angiography is an invasive method and it should be the first step to endovascular treatment. CT angiography is a noninvasive method, which can be used for diagnosis. Moreover, endoscopy was suboptimal in our case because blood was found without the bleeding site. Small metalic clips left in the bleeding site are very helpful in finding the target for endovascular treatment. Visceral aneurysms are the hallmark of PAN, with a reported sensitivity and specificity of 89% and 90%, respectively [8]. Percutaneous transcatheter embolization has been used with increasing frequency in the management of ruptured visceral artery aneurysms associated with PAN. Coils and glue are used for embolization of microaneurysms in PAN. In our case, we preferred to use glue as an embolic agent and were able to embolize the bleeding vessel successfully.

Conclusions

To the best of our knowledge, this is the youngest child reported on, with massive GI bleeding secondary to PAN, treated with successful percutaneous transcatheter embolization under emergency conditions. Angiography in such cases can be useful for endovascular treatment of gastrointestinal bleeding resulting from ruptured microaneurysms.

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