



**Case Report**

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# Angiodysplasia of the Ileum: Case Report of a Rare Cause of Intestinal Bleeding in Children



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## Abstract

**Introduction:** Angiodysplasia can occur throughout the gastrointestinal tract and ileum is one of the rarest locations for angiodysplasias in the overall population. There may be multiple angiodysplasias in one gastrointestinal part or they can coexist in several different gastrointestinal location. Gastrointestinal angiodysplasia is most often founded in patients older than 60 years.

**Case presentation:** This is a case about a 14 year old Croatian white female who initially presented with symptoms of syncope and iron deficiency anemia. Scintigraphy with Technetium (TC)- 99m pertechnetate revealed abnormal focal activity in a segment of the small intestine in the right abdominal part and we believed that this was due to a Meckel's diverticulum. During the operation we found an angiodysplasia of the ileum.

**Conclusion:** This case highlights the importance of considering a gastrointestinal angiodysplasia in an acute pediatric gastrointestinal bleeding patient and as a differential diagnosis of anemia. During an operation, children with gastrointesinal bleeding require careful examination of the gastrointestinal tract and should be carried out in all cases where an initial laparoscopy failed to identify a cause of bleeding.

**Key words:** Angiodysplasia; Vascular malformation; Gastrointestinal hemorrhage; Ileum; Children; Case report

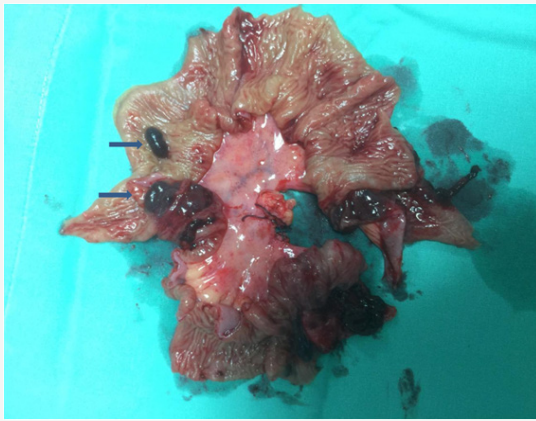
## Introduction

The prevalence of gastrointestinal angiodysplasia in the overall population is not well known. In the western population the jejunum was the most common location of angiodysplasias (80%), duodenum (51%), stomach (22.8%), right colon (11.4%) and ileum (5.7%) [1]. There are a few cases in the literature about gastrointestinal angiodysplasias in the children [2-4]. Histological confirmation shows dilated vessels in the mucosa and submucosa, sometimes covered with single layer of surface epithelium [5].

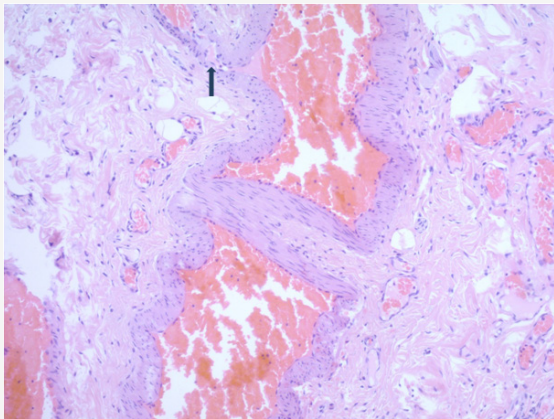
## Case presentation

A 14 year old Croatian white female presented with a history of syncope and iron deficiency anemia. She had juvenile arthritis and she was treated with methotrexate, methylprednisolone, meloxicam and acidum folicum. She was pale and hemodynamically stable. Her hemoglobin was 63g/L, red blood cells was  $2.2 \times 10^{12}/L$ , Fe  $2 \mu\text{mol}/L$ , and had a positive fecal occult

blood. Scintigraphy with Technetium (TC)- 99m pertechnetate and SPECT (single photon emission computed tomography) abdominis showed activity in the stomach and abnormal focal activity in a segment of the small intestine in the right abdominal part and we believed that this was due to a Meckel's diverticulum. During laparoscopic examination we could not find any source of bleeding so we proceeded with a laparotomy. We saw blood in the lumen of ileum and three lesions were identified on palpation of the small intestine at approximately 90cm. orally from the valvulae Bauhini. A 25cm portion of ileum was resected and a primary anastomosis was performed (Figure 1). Hystological findings were consistent with angiodysplastic lesions, there were a multiple tortuous thin-walled vessels in submucosa with anastomosis between vessels (Figure 2). She recovered well and was discharged on seventh post operative day. During a one year follow up she was without symptoms such as rebleeding or anemia.



**Figure 1:** Gross photograph of resected ileum.



**Figure 2:** Angiodysplasia of the ileum shows irregular dilatation and thinning of submucosal arteries and veins. A-V anastomosis (arrow) HE x 100.

## Discussion

Angiodysplasia account for 60%-75% of bleeding sources in the small bowel and are generally found in the proximal small bowel [1,6]. Exact data is not known for children yet. We found sources of bleeding in ileum which is the rarest location for angiodysplasias. The bleeding in angiodysplasia tend to be recurrent and chronic [7]. Nevertheless, profuse acute bleeding can be the reason for orthostasis and hypotension. Almost 50% of patients presenting with anaemia are found to have angiodysplasia [8]. In our case the first clinical manifestation was syncope and iron deficiency anemia but we seldom think about angiodysplasia in the children. Sometimes, the diagnostic examinations of causes of bleeding from the gastrointestinal are very difficult and inaccurate. Angiodysplasia is usually diagnosed by endoscopy, capsule endoscopy, MSCT or MR angiography and standard angiography [1,6,9,10]. In one serie selective viscerale angiography was the- most valuable investigation, identifying the probable cause of bleeding in 53% of all patients [8]. Angiography is an invasive diagnostic tool, has many complications, is dependent on the quality of angiography and experience of the physician carrying out the radiological interpretation. Sometimes

surgery may be required for detection of angiodysplasias. In one serie, exploratory laparotomy detected 21% of identified causes of bleeding and was particularly valuable in younger patients. Other examination methods such as small bowel radiography and radionuclide bleeding scan are not satisfactory for detecting small bowel lesions [6]. In our case, a radionuclide scan was misleading. We believed that the source of bleeding was from a Meckel's diverticulum. During laparoscopy we failed to find a Meckel's diverticulum nor the source of bleeding. We proceeded with a laparotomy and performed a careful examination from the stomach to the rectum and found multiple angiodysplasias in the ileum. Treatment angiodysplasia ranged from observation to endoscopic treatment, medications, laser or surgery. The rebleeding rate after endoscopic therapy for angiodysplasia of the small bowel is about 45% [11]. Surgical resection is the method of choice for lesions that have been clearly identified as the source of severe bleeding and for multiple lesions with recurrent bleeding [1,3,6,8,9]. Failing to find a source of bleeding can be the cause of death. In patients who experience symptomatic blood loss due to angiodysplasia, approximately 40-50% will experience rebleeding [12]. So we need to identify these lesions and perform a definitive treatment. We resected part of the ileum that had angiodysplasia and a primary anastomosis was performed which proved to be a safe choice.

## Conclusion

This case highlights the importance of considering a gastrointestinal angiodysplasia in an acute pediatric gastrointestinal bleeding patients and during diagnostic of anemia. Scintigraphy with Technetium (TC)- 99m pertechnetate and SPECT abdominis can't distinguish between Meckel's diverticulum and angiodysplasia. During an operation in children with gastrointestinal bleeding, a careful examination of the gastrointestinal tract should be carried out in all cases where an initial laparoscopy failed to identify a cause of bleeding. Resection of the bowel with a primary anastomosis is a definitive therapy for angiodysplasia.

## Compliance with Ethical Standards

### Ethical approval

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

### Informed consent

Informed consent was obtained from all individual participants included in the study.

### Conflict of Interest

Author Stanko Čavar, Author Mate Škegro, Author Dražen Jelašić, and Author Jurica Vuković declare that they have no conflict of interest.

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