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, methylprednisolonom, mekoxicamum and acidum folicum. She was pale and hemodynamically stable. Her hemoglobin was 63g/L, red blood cells was 2.2 x 1012/L, Fe 2 umol/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 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 gastrointestinal 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
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 angiodisplasia 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 visceral 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 exprience 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 Meckels' 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 treament, 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 angiodyplasia 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 anaemia. 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 definiteve 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 Ćavar, Author Mate Škegro, Author Dražen Jelašić, and Author Jurica Vuković declare that they have no conflict of interest.
References


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