

Colorectal Lymphangioma Circumscriptum in Child: A Rare Case Report

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Abstract: ***Background:** Lymphangioma is a rare malformation in the lymphatic system. Lymphangioma generally occurs in the neck and axilla, only 1% of the cases are found in the retroperitoneal organ. We reported a rare case of colorectal lymphangioma circumscriptum in a 4 years old child and colonoscopy was performed. **Case Presentation:** A 4 years old boy came with intermittent hematochezia since 2 years old, periumbilical abdominal pain, nausea, vomiting, constipation and a mass protruding from the anus after defecation for 2 days duration. The patient was initially diagnosed with a suspected colorectal polyp, then polypectomy via colonoscopy was performed. Histopathological examination of colonic tissue revealed a lymphangioma circumscriptum in the descending colon. **Discussion:** Lymphangioma can be diagnosed through a detailed history taking, physical examination, and histopathological examination. The role of colonoscopy and histopathological in colorectal lymphangioma is quite significant, as it is mostly performed for diagnosis. Excision of colorectal lymphangioma is required to prevent complications. **Conclusion:** Colorectal lymphangioma circumscriptum is a congenital benign tumor originating from the lymphatic system with a variety of digestive tract symptoms. The widely used diagnostic modality in this case is colonoscopy. Surgical removal of neoplasms is the best option for treating lymphangiomas.*

Keywords: colorectal lymphangioma circumscriptum, colonoscopy

1. Introduction

Lymphangioma is a rare malformation in the lymphatic system. Lymphangioma can be found on the skin and mucous membranes.^{1,2} It is classified into deep lymphangioma which is usually congenital and superficial lymphangioma which is mostly acquired except for the circumscriptum type which appears from birth.^{2,3} Lymphangioma generally occurs in the neck and axilla, only 1% of the cases are found in the retroperitoneal organ.^{4,5} We reported a rare case of colorectal lymphangioma circumscriptum in a 4 years old child and colonoscopy was performed.

2. Case Presentation

A 4 years old boy was admitted to Siloam Hospital Bangka Belitung with intermittent hematochezia since 2 years ago and there was a mass protruding from the anus after defecate since 2 days prior to admission. Periumbilical abdominal pain, nausea, vomiting, and constipation were found. The patient denied weight loss, but inability to gain weight in the past 2 years. The patient's current weight is 13.67 kg and categorized as underweight based on age. The patient also had a history of cow's milk allergy. On physical examination, there was a single mass (approximately 1 cm) protruding from the anus with a smooth surface and easily bled. Laboratory examination was performed: Hb 11.4 g/dL, leukocyte 5.000/ μ L, platelet 340.000/ μ L, and positive occult blood test.

The patient was initially diagnosed with a suspected colorectal polyp with a differential diagnosis of colorectal

tumor, then a colonoscopy examination and polypectomy via colonoscopy or laparoscopic colon resection was planned if multiple colon polyps was found. Yet, only a colonoscopy was performed. There was dilation of the blood vessels in the inferior hemorrhoid plexus. The rectum and the descending colon with polypoid masses were visualized measuring approximately 0.5cm and 2cm respectively. The masses were then removed and a histopathological examination was carried out. An acutely inflamed (suppurative) necrotic tissue was found on rectal tissue histopathology, while histopathological examination of colonic tissue revealed a tumor with a single layer of columnar epithelium and several crypts in the subepithelium consisted of a proliferation of lymph vessels without any signs of malignancy. These findings conclude a lymphangioma circumscriptum in the descending colon. After the surgery, there were no complaints of hematochezia and the patient was able to defecate normally.



Figure 1. Colonoscopy performed at our hospital revealed a mass finding in the descending colon

3. Discussion

Lymphangioma is a rare tumor of the lymphatic system that is generally benign and occurs either congenitally or acquired. Based on its size and extension, lymphangioma is classified into deep and superficial lymphangioma. Deep lymphangiomas such as cavernous and cystic lymphangiomas were found congenital, while superficial lymphangiomas can present either congenital or acquired. lymphangioma circumscriptum is a superficial lymphangioma that is generally found at birth.^{1,2,5}

Lymphangioma can be found on the skin and mucous membranes of any part of the body, mostly in the neck (75%) and armpits (15%). Colorectal lymphangioma is an infrequent disease, its incidence is only 1%.^{4,5} Colorectal lymphangioma generally occurs after the age of 40 years and is more common in men than women, while in this case it is diagnosed in a child.⁵ Several risk factors accounted for colorectal lymphangioma are history of inflammation of the gastrointestinal tract, surgical procedures, and radiation, whereas in this case no specific risk factors were identified.⁵ Colorectal lymphangioma can manifest with or without digestive tract symptoms such as abdominal pain, bloody stools, diarrhea, or constipation. In this case, the patient experienced digestive tract symptoms (abdominal pain, nausea, vomiting, bloody stools, and constipation).⁵

Lymphangioma can be diagnosed through a detailed history taking, physical examination, and histopathological examination.⁶ Lymphangioma can be macrocytic (>1cm), microcytic (<1cm), or both.⁴ Lymphangioma circumscriptum commonly appears as multiple tumors, either in groups or spread, characterized by hemorrhagic vesicular papules as they consist of a combination of vascular and lymph vessels.² The role of colonoscopy in colorectal lymphangioma is quite significant, as it is mostly performed for diagnosis.^{5,7} In our colonoscopy findings, there were multiple hemorrhagic lesions and ungrouped in the descending colon and rectum. On histopathological examination of colorectal lymphangioma, there was a large proliferation of lymph vessels in the tissue and this will reduce the number of crypts in the lamina propria.^{7,8}

Surgical removal of neoplasms is the best option for treating lymphangiomas.¹ The success rate of surgery is higher in cases of superficial and small lymphangiomas. Wide local excision is needed because of the high recurrence rate (up to 23% in the 81 months after excision), thus regular check-up is needed in lymphangioma patients.^{2,9} In our case, the patient denied any symptoms after 20 months of evaluation post-colonoscopy.

4. Conclusion

Colorectal lymphangioma circumscriptum is a congenital benign tumor originating from the lymphatic system with a variety of clinical manifestations, especially hematochezia, and the case is infrequent in the colorectal region. The widely used diagnostic modality in this case is colonoscopy. Excision of colorectal lymphangioma is required to prevent complications and the necessity of histopathological

examination is to confirm diagnosis and to exclude the possibility of malignancy.

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