CASE REPORT

COLONIC MALAKOPLAKIA

Arit Prakash[1], Noshaba Noor[1], Ayesha Marchant[1], Mariam Ghori[2], Rameen Akhtar Molani[2]

[1]National Institute of Child Health, Karachi-Pakistan
[2]Jinnah Postgraduate Medical Centre, Karachi-Pakistan

This case report is of a 10-year-old female who presented with complaints of blood in her stool and colicky abdominal pain for the last 1.5 years. Biopsy samples from the rectum and left-sided colon were taken during a colonoscopy, revealing numerous histiocytes having calcified bodies (Michaelis-Gutmann bodies). Diagnosis of Malakoplakia was confirmed with no evidence of dysplasia, malignancy, or granuloma.

Keywords: Malakoplakia; Abdominal pain; Michaelis-Gutmann bodies

Citation: Prakash A, Noor N, Marchant A, Ghori M, Molani RA. Colonic Malakoplakia. J Ayub Med Coll Abbottabad 2024;36(2):451–3.

DOI: 10.55519/JAMC-02-12643

INTRODUCTION

Malakoplakia is a rare granulomatous inflammatory disorder. It is especially rare in the pediatric age group.[1] In a recent case series of 23 patients with Malakoplakia of the gastrointestinal tract, the mean age recorded was 57 years.[2] Unfortunately, statistical data on the incidence or prevalence of colonic malakoplakia in pediatric patients is limited due to its rarity. Its presentation depends on the organ system involved.[1] After the genitourinary tract, the gastrointestinal tract is the most common system involved. In the GIT, the rectum and sigmoid colon are the most frequently affected sites.[2,3] Other sites may be involved, such as the pancreas, liver, lymph node, skin, respiratory tract, vagina, and skin.[1] Malakoplakia can present in various ways, the most common symptoms being abdominal pain, fever, diarrhea, and rectal bleeding.[3] The disease is diagnosed based on histopathology findings of numerous histiocytes having calcified bodies known as Michaelis Guttman bodies.[4] Considering the severity of the clinical presentation, the treatment ranges from antibiotic therapy to surgery.[3] The importance of reporting this case report is to highlight that when patients present with abdominal pain and bloody diarrhea they should be thoroughly investigated. If the symptoms persist for weeks or months, invasive procedures like colonoscopy with biopsy should be done as histopathology will help diagnose these cases.

CASE PRESENTATION

This case report explains a case of a 10-year-old female weighing 20 kgs who presented to us with complaints of blood in stool and colicky abdominal pain in her left lower abdomen, insidious in nature, relieved with defecation and associated with tenesmus. There was no radiation of pain, and it occurred throughout the day. Abdominal pain was associated with on and off loose motions around a cup in quantity mixed with mucus and often with blood, too, for the last 1.5 years.

She went to local doctors, where blood tests, stool tests, and other investigations were done. Her stool and blood cultures tested negative. Her histopathology report of section of colonic mucosa showed preserved architecture. No cryptitis, mucin depletion or crypt abscess is noted. One of the fragments shows numerous histiocytes having calcified bodies (Michaelis-Gutmann bodies), indicating Colonic Malakoplakia. No evidence of dysplasia, granuloma or malignancy is seen. No cryptitis, mucin depletion or crypt abscess is noted.

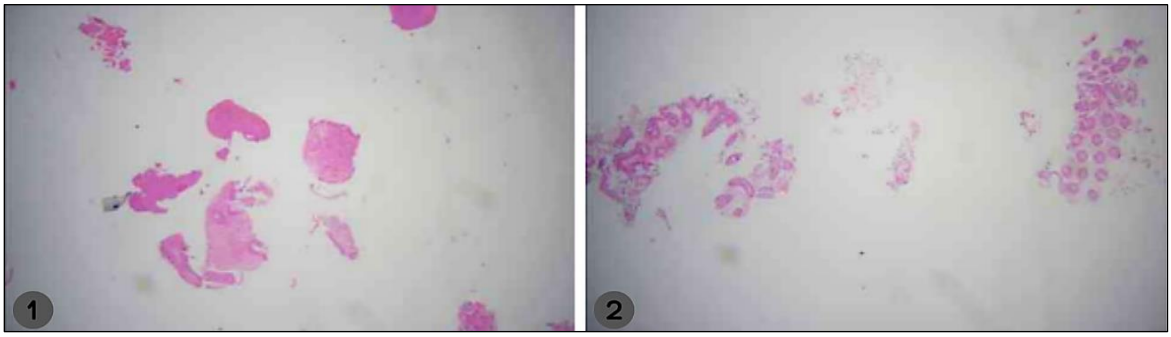

Fig 1. and Fig 2. Photomicrograph shows fragments of mucosa from rectum (left sided colon) and splenic flexure. One of the fragments from left sided colon shows numerous histiocytes having calcified bodies (Michaelis-Gutmann bodies)

Department of Histopathology

Biopsy No.	S22-45706
Specimen:	MUCOSA FROM RECTUM (LEFT SIDE COLON) AND SPLENIC FLEXURE
Clinical Findings:	**- Abdominal pain and loose stools for 1.5 years**
Gross:	Specimen is received in formalin in two containers: **A): Mucosa from rectum (left side colon):** The specimen consists of five mucosal fragments measuring 0.5x0.5x0.2cm in aggregate. Specimen is submitted entirely in one block. **B):Splenic flexure:** The specimen consists of four mucosal fragments measuring 0.3x0.3x0.2cm in aggregate. Specimen is submitted entirely in one block.
Microscopy:	A): Histological examination of section reveals colonic mucosa with preserved architecture. No cryptitis, mucin depletion or crypt abscess is noted. One of the fragment shows numerous histiocytes having calcified bodies (Michaelis-Gutmann bodies). No evidence of dysplasia, granuloma or malignancy is seen. B): Histological examination of the sections from large bowel mucosa reveal preserved architecture. No cryptitis, crypt abscess, crypt dropout or mucin depletion. No evidence of granuloma or malignancy is seen.
Diagnosis:	**A): MUCOSA FROM RECTUM (LEFT SIDE COLON):** **- Malakoplakia** **- No evidence of granuloma or malignancy is seen** **B): SPLENIC FLEXURE:** **- Unremarkable Mucosa With No Significant Pathology** **- No evidence of granuloma or malignancy is seen**

She was treated with oral antibiotics accordingly and was kept on metronidazole each time by a local general physician for around five to seven days. She was given either metronidazole or metronidazole + nitrofurantoin combinations alternatively. She was even given Intravenous (IV) medications, but no permanent improvement was noticed. She was then referred to the pediatric gastroenterology department National Institute of Child Health, Karachi (NICH).

On physical examination at NICH, she was weak, pale, and lethargic, and her mother reported low appetite and weight loss. There was no associated history of fever, joint pain, jaundice, or oral ulcers. Moreover, no hair loss, red eye, photophobia, or skin rash was observed. Also, there was no complaint of perianal pain, itching, or discharge. However, she had alternating bowel habits.

There was no history of hospital admission or blood transfusion. Similarly, she had no previous surgical history too. Her birth history was unremarkable. Her diet included homemade food, approximately 1300 kcal/day. She was developmentally appropriate for her age and vaccinated, according to EPI. She is the 3rd alive child of her consanguineously related parents, and her other siblings are healthy and alive. No history of Tuberculosis (TB) contact was given. They used boiled water and belonged to an overall low socioeconomic status.

A CT scan abdomen was done at our setup NICH, which suggested circumferential thickening of the rectum, the rectosigmoid junction (Fig1-5), and the sigmoid colon. Also, there was a thickening of the ascending and descending colon. Findings were most likely suggesting inflammatory bowel disease. Keeping the persistent symptoms in view, her colonoscopy was planned. Biopsy samples from the rectum and left-sided colon were taken during a colonoscopy, revealing numerous histiocytes having calcified bodies (Michaelis-Gutmann bodies). Malakoplakia was diagnosed with no evidence of dysplasia, malignancy, or granuloma. (Fig. 1-5) Oral ciprofloxacin 10 mg/kg/dose twice a day has been prescribed to the patient and the patient's family was counseled regarding surgical resection. However, the patient was lost to follow-up.

CASE DISCUSSION

Malakoplakia is a rare granulomatous inflammatory disease, especially in the pediatric age group.[1] Based on the clinical presentation, it is not diagnosed and,

therefore, not timely treated or documented in Pakistan.

Our case report focuses on a rare presentation of Malakoplakia in Pakistan in the pediatric age group, where a ten-year-old female presented with recurrent diarrhea containing mucus and blood. She was treated symptomatically with antibiotics on every visit to the local hospitals, with inflammatory bowel disease kept as a differential. She was not diagnosed with Malakoplakia for an extended time due to a lack of awareness of Malakoplakia among the local doctors. In Pakistan, diseases are often treated symptomatically, and their underlying causes are not being identified or considered as differential due to limited resources and inadequate research opportunities or knowledge among the local doctors.

Malakoplakia is mainly diagnosed based on histopathology findings of histiocytes having numerous calcified bodies known as Michaelis-Gutmann bodies.[4] Similarly, at our setup in NICH, this patient was diagnosed. She presented with weight loss, recurrent left lower abdominal pain associated with tenesmus, on and off loose stools mixed with mucus and often blood, which directs towards the involvement of the gastrointestinal tract. Later her Computed tomography (CT) scan of the abdomen was done, which showed thickening of the rectum, rectosigmoid junction, sigmoid colon, and ascending and descending colon, pointing towards Inflammatory Bowel disease. On further investigation, her colonoscopy was done, which revealed numerous histiocytes having calcified bodies (Michaelis-Gutmann bodies) on the histopathologic report of the biopsy. The diagnosis of Malakoplakia of the gastrointestinal tract was completed with no evidence of dysplasia, malignancy, or granuloma.

In addition, since Malakoplakia is a rare disease, there's no standard treatment guideline for it worldwide.[3] However, antibiotics with or without surgical resection are the only supportive treatment.[3]

The primary motive of this case report is to draw the attention of doctors in developing countries, specifically Pakistan, where Malakoplakia is very rare. Doctors don't consider Malakoplakia of the gastrointestinal tract as a differential when patients present with recurrent abdominal pain, loose stool, and weight loss for an extended time. Early diagnosis and timely management may prevent further complications and improve the prognosis of Malakoplakia.

CONCLUSION

Malakoplakia is a rare inflammatory granulomatous disease involving various systems, second most commonly gastrointestinal system. The symptoms may vary from abdominal pain to rectal bleeding and weight loss. Diagnosis is based on histopathology showing Michaelis-Gutmann bodies. Therefore, in developing countries such as Pakistan, with limited resources in the local hospitals and inadequate knowledge regarding Malakoplakia among the local doctors, colonoscopy, and histopathology should be considered to rule out Malakoplakia upon clinical presentation for timely diagnosis and treatment. Moreover, there is a further need for research on the standard treatment guidelines for Malakoplakia.

REFERENCES

1. Jadhav MN, Patil RS, Kittur SK, Yadav J. Colorectal malakoplakia in a child presenting as multiple polyposis coli. Indian J Pathol Mircrobiol 2015;58(1):86–6.
2. Lee M, Ko HM, Rubino A, Lee H, Gill R, Lagana SM. Malakoplakia of the gastrointestinal tract: clinicopathologic analysis of 23 cases. Diagn Pathol 2020;15(1):97.
3. Achufusi TG, Jessamy K, Chebaya P, Rawlins S. Rectal malakoplakia. Proc (Bayl Univ Med Cent). 2020;33(3):389–90.
4. Yared Rawad A, Badran Hussein A, Kamareddine M, Ghosn Y, Bou Khalil R, El Ajamy K, *et al*. Colonic Malakoplakia: A Rare Finding in a Healthy Male. Case Rep Gastroenterol 2018;12(2):453–6.

Submitted: November 13, 2023	*Revised: March 19, 2024*	*Accepted: April 25, 2024*

Address for Correspondence:

Dr. Rameen Akhtar Molani, Jinnah Postgraduate Medical Centre, Karachi-Pakistan
Cell: +92 342 269 8002
Email: Rameenakhter@hotmail.com