

What Lies Beneath – Malakoplakia : A Rare Submucosal Lesion of the Colon

ABSTRACT

Malakoplakia is a rare multiorgan granulomatous disease that can be found within the gastrointestinal tract. Its appearance can vary including polyps, ulcerations, plaques, flat lesions, erosions or even large mass lesions. Given its varied endoscopic appearance there exists a broad differential diagnosis including malignancy or pre-malignant neoplasm. Malakoplakia within the gastrointestinal tract has been observed in coexistence with colorectal adenocarcinoma; however, no direct causal association has been identified. Given the possible malignant differentials of colonic lesions, a histologic assessment is required for the diagnosis of malakoplakia. Malakoplakia is generally considered a benign condition with no specific follow-up required in the gastrointestinal tract. We present a case of a rare submucosal finding of malakoplakia in the ascending colon discovered on colonoscopy.

Keywords: Malakoplakia, Submucosal, Colon, Gastrointestinal

1. INTRODUCTION

A variety of submucosal lesions can be found in the colon and are often an incidental finding. Most are benign including lipomas, leiomyomas, and lymphoid aggregates. Malignant causes also exist including neuroendocrine tumours and metastatic lesions. Soft lesions generally represent lipomas, confirmed by a positive "pillow sign". However, accurate diagnosis of a firm submucosal lesion requires histologic assessment. We present a case of a rare benign firm submucosal lesion discovered on colonoscopy.

2. PRESENTATION OF CASE

A 53-year-old female underwent gastroscopy and colonoscopy for investigation of iron deficiency anaemia. There were no gastrointestinal symptoms or history of weight loss. There was no history of immune suppression, autoimmune disease, or chronic infection. Gastroscopy was unremarkable. At colonoscopy, one 3mm firm submucosal nodule was found in the proximal ascending colon (Figure 1 Panel A/B). This was resected en-bloc using a cold polypectomy technique. No other abnormalities were found.

Histology demonstrated a well-defined lesion in the mucosa and submucosa composed of abundant histiocytes admixed with small numbers of lymphocytes, plasma cells, neutrophils and rare eosinophils. Many of the histiocytes contain characteristic Michaelis-Gutmann bodies which are Periodic acid-Schiff positive (Figure 1 Panel C/D).

3. DISCUSSION

Malakoplakia is a rare granulomatous disease that can involve many organ systems. The exact pathogenesis is poorly understood but is thought to be related to impaired macrophage ability to phagocytose bacteria leading to formation of pathognomonic Michaelis-Gutmann bodies [1]. This dysfunction is hypothesised to be related to abnormalities in the lysosomal system within macrophages. Organisms isolated from malakoplakia lesions include escherichia coli, klebsiella pneumoniae, mycobacterium tuberculosis, proteus, rhodococcus equi, staphylococcus aureus and pseudomonas aeruginosa [2]. There has been an association of malakoplakia with conditions of immune dysregulation [2].

Malakoplakia is most commonly seen in the genitourinary tract followed by the gastrointestinal tract. Less commonly it is also reported to involve the pancreas, lymph nodes, central nervous system, middle ear, tongue, tonsils, conjunctiva, thyroid, bone, skin, prostate, breast, adrenal and respiratory tract [2, 3]. Gastrointestinal malakoplakia, while the second most common site of involvement, remains an uncommon entity with limited cases reported in the literature, likely due to under recognition and underreporting. The diagnosis can be challenging due to its nonspecific clinical presentation and rarity. Patients can present with symptoms of abdominal pain, diarrhoea and haematochezia, however in many cases the lesion is

asymptomatic and discovered incidentally like in our case [3, 4]. In spite of what its name suggests (derived from the Greek “malakos” soft and “plakos” plaque), the endoscopic appearance can vary including polyps, ulcerations, plaques, flat lesions, erosions or even large mass lesions [3, 4]. Malakoplakia has been identified from the stomach to the anus though most commonly observed in the sigmoid colon and rectum [3]. Given its varied endoscopic appearance there exists a broad differential diagnosis including malignancy or pre-malignant neoplasm.

Malakoplakia in the genitourinary tract has been reported in coexistence with urothelial carcinoma, MALT lymphoma, and prostate cancer [1,4]. Within the gastrointestinal tract, malakoplakia has been observed a coexistence with colorectal adenocarcinoma however no direct causal association has been identified [5]. It has been hypothesised that coincidence of adenocarcinoma with malakoplakia may be a result of the distortion of the local microbiota by the malignancy [6]. Malakoplakia is generally considered a benign condition with no specific follow-up required in the gastrointestinal tract. Given its coexistence with colorectal cancer, would warrant a careful search for colorectal cancer.

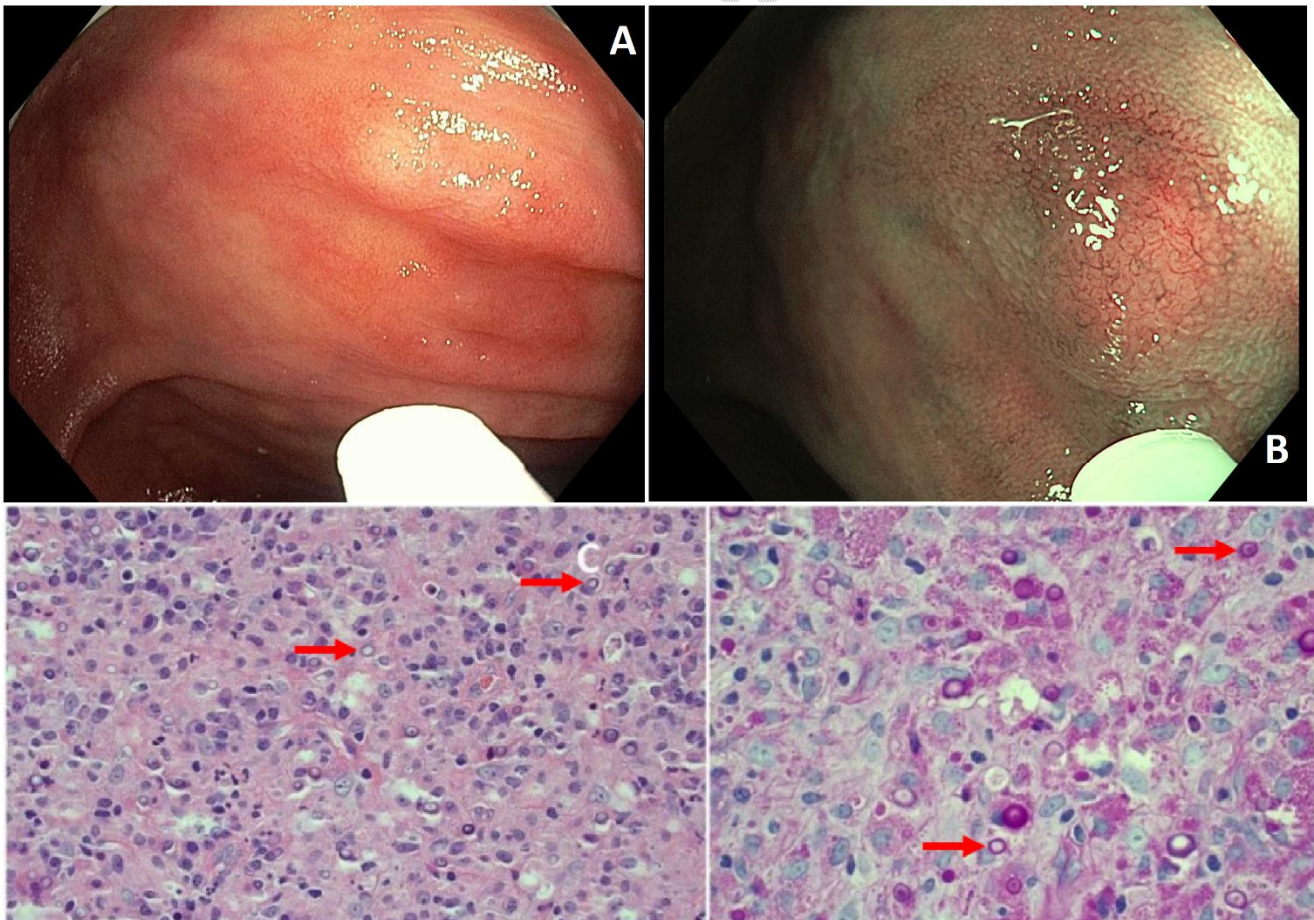


Fig.1. Panel A/B: White light and NBI image of submucosal nodule found in the ascending colon Panel C/D: Periodic acid-Schiff (x40) stain with abundant eosinophilic granular cytoplasm and numerous intracellular Michaelis-Gutmann bodies

4. CONCLUSION

Given the possible malignant differentials of colonic lesions, a histologic assessment is required for the diagnosis of malakoplakia. Malakoplakia is generally considered a benign condition with no specific follow-up required in the gastrointestinal tract. Given its coexistence with colorectal cancer, would warrant a careful search for colorectal cancer.

Disclaimer (Artificial intelligence)

Option 1:

Author(s) hereby declare that NO generative AI technologies such as Large Language Models (ChatGPT, COPILOT, etc) and text-to-image generators have been used during writing or editing of manuscripts.

Option 2:

Author(s) hereby declare that generative AI technologies such as Large Language Models, etchave been used during writing or editing of manuscripts. This explanation will include the name, version, model, and source of the generative AI technology and as well as all input prompts provided to the generative AI technology

Details of the AI usage are given below:

- 1.
- 2.
- 3.

CONSENT

As per international standards or university standards, patient(s) written consent has been collected and preserved by the author(s).

ETHICAL APPROVAL (WHEREEVER APPLICABLE)

Ethics approval was gained from the Metro South Human Research Ethics Committee.

REFERENCES

- [1] Cieżczyk K, Puderecki M, Wronecki L, et al. Malakoplakia of the urinary system. Folia Med Cracov. 2019;59(2):67-74.
- [2] Yousef GM, Naghibi B, Hamodat MM. Malakoplakia outside the urinary tract. Arch Pathol Lab Med. 2007 Feb;131(2):297-300. doi: 10.5858/2007-131-297-MOTUT. Erratum in: Arch Pathol Lab Med. 2009 Jun;133(6):850
- [3] Lee M, Ko HM, Rubino A, Lee H, Gill R, Lagana SM. Malakoplakia of the gastrointestinal tract: clinicopathologic analysis of 23 cases. DiagnPathol. 2020 Jul 24;15(1):97.

- [4] Matsuda I, Zozumi M, Tsuchida YA, et al. Primary extranodal marginal zone lymphoma of mucosa-associated lymphoid tissue type with malakoplakia in the urinary bladder: a case report. *Int J ClinExpPathol.* 2014 Jul 15;7(8):5280-4.
- [5] Andrés L, Etxegarai L, López JI, et al. Malakoplakia associated with colorectal adenocarcinoma. *Ann Saudi Med.* 2005;25(1):50-52.
- [6] Bates AW, Dev S, Baithun SI. Malakoplakia and colorectal adenocarcinoma. *Postgrad Med J.* 1997 Mar;73(857):171-3. doi: 10.1136/pgmj.73.857.171.
- [7] Rubio CA. Colorectal Carcinogenesis from Gut-associated Lymphoid Tissue Clinical and Experimental Documentation. *J. Adv. Med. Med. Res.* [Internet]. 2017 Apr. 26 [cited 2024 May 24];21(1):1-12. Available from: <https://journaljammr.com/index.php/JAMMR/article/view/3199>
- [8] Ranbhare A, Ali SF, Jadhav S. Adult Colocolic Intussusception Secondary to Lipoma – Enterotomy over Resection—A Different Approach. *Asian J. Case Rep. Surg.* [Internet]. 2023 Mar. 31 [cited 2024 May 24];6(1):123-9. Available from: <https://journalajcrs.com/index.php/AJCRS/article/view/380>
- [9] Fujiya M, Tanaka K, Dokoshi T, Tominaga M, Ueno N, Inaba Y, Ito T, Moriichi K, Kohgo Y. Efficacy and adverse events of EMR and endoscopic submucosal dissection for the treatment of colon neoplasms: a meta-analysis of studies comparing EMR and endoscopic submucosal dissection. *Gastrointestinal endoscopy.* 2015 Mar 1;81(3):583-95.