CASE REPORT

Rectosigmoid malakoplakia

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SUMMARY

We present a rare case of rectosigmoid malakoplakia treated with fluoroquinolone.

BACKGROUND

Malakoplakia is a granulomatous inflammation that involves the urogenital tract, gastrointestinal tract, skin, lungs and the central nervous system. It presents a diagnostic challenge as clinical presentation can be non-specific. We report a case of a middle-aged woman who presented with diarrhoea and was diagnosed with malakoplakia.

CASE PRESENTATION

A 61-year-old woman with a medical history of diabetes mellitus, hypertension and hyperlipidaemia was admitted for lower respiratory tract infection. She also reports three to five bowel movements daily for 6 months associated with subjective weight loss. General physical and abdominal examinations were unremarkable.

INVESTIGATIONS

Laboratory investigations revealed microcytic and hypochromic anaemia (mean corpuscular volume 79.4 fL, mean corpuscular haemoglobin 25.1 pg, mean corpuscular haemoglobin concentration 31.5 g/dL) with haemoglobin 10.0 g/dL. White blood cell count was $10.4 \times 10^9/L$ with 25.2% lymphocytes. She was iron deficient (ferritin 10 µg/L, transferrin 244 ng/dL, total iron binding capacity 63 µmol/L, iron saturation 8%). HIV antigen-antibody screen was negative and HbA1c was 10.9%.

CT scan of the thorax, abdomen and pelvis was unremarkable.

Colonoscopy showed multiple colonic polyps. The largest polyps were seen in the distal sigmoid colon. These polyps were sessile and snared. Histology confirmed these polyps as serrated adenoma without dysplasia. A cluster of inflammatory polyps with ulcerative edges were noted in the rectosigmoid region (figure 1). Histology of these polyps showed inflammatory infiltrate with prominent histiocytic component without well-formed granulomas or necrosis. There were several Michaelis-Gutmann bodies (positive for von Kossa and Perl's stain) seen (figures 2, 3 and 4). No fungal organisms or acid fast bacilli were evident on Grocott's methenamine silver and Ziehl-Neelsen stains, respectively. Diagnosis of malakoplakia was established.

DIFFERENTIAL DIAGNOSES

1. Tuberculosis
2. Fungal infections
3. Colonic malignancy
4. Lymphoma

TREATMENT

We treated our patient with 3 weeks of oral ciprofloxacin.

OUTCOME AND FOLLOW-UP

Patient is well on a follow-up telephone conversation. She subsequently underwent a repeat flexible sigmoidoscopy which revealed persistent cluster of polyps at rectosigmoid junction. Repeat biopsies revealed focal features of hyperplastic polyp with no evidence of malakoplakia seen.

DISCUSSION

Malakoplakia is a rare chronic inflammation that is characterised microscopically by the accumulation of tissue macrophages containing unique intracytoplasmic calcified inclusion bodies, known as Michaelis-Gutmann bodies. Various Gram-negative organisms have been reported to cause malakoplakia, most commonly *Escherichia coli*. The most common site of involvement is the urogenital tract followed by the gastrointestinal tract. Within the gastrointestinal tract, the rectum is the most common site followed by the sigmoid colon.¹

Malakoplakia of the colon may cause symptoms such as abdominal pain, diarrhoea, rectal bleeding and fever.² Malakoplakia presents in the early stages as soft yellow to tan mucosal plaques or, in the late stages, as raised, grey to tan lesions in various sizes with peripheral hyperaemia and a central depressed area.³ However, none of these symptoms are characteristic, and due to the rarity of the disease, it is often incidental. In addition, malakoplakia can mimic malignancy. This makes diagnosis of malakoplakia a challenge and is usually diagnosed histopathologically.

Malakoplakia is more commonly reported in patients with immunocompromised states such as post organ transplantation, diabetes mellitus (as in our patient), AIDS, malignancy (commonly colon carcinoma) and immunosuppressive therapy.⁴ It is also associated with chronic *E. coli* infection.⁵ These associations suggest a malfunction of T lymphocytes involved in the pathogenesis of malakoplakia.

Due to the rarity of the condition, there are no standard treatment guidelines. However, mainstay of treatment are antibiotics that are concentrated in macrophages, that is, quinolone

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Figure 1 Colonoscopic image showing rectosigmoid cluster of polyps.

and trimethoprim-sulfamethoxazole, as well as treating the underlying immunosuppressive condition. *Ugeskr Laeger* have reported successful treatment of colonic malakoplakia with 3 months of sulfamethizole and trimethoprim.⁶ Hyun *et al* have also reported successful treatment of rectal sigmoid malakoplakia with 6 months of ciprofloxacin, the cholinergic agonist bethanechol and a multivitamin.⁷ Ascorbic acid has also been suggested to be effective.^{8,9} Duration of the antibiotic therapy reported is variable, depending on the location/extent of infection and if there are complications, for example, abscess formation. Based on the outcome of our case, we reckon that 3 weeks

Figure 2 Polypoid large bowel mucosa with expanded lamina propria (H&E low-power magnification).

Figure 3 The expanded lamina propria contains some lymphocytes and abundant macrophages (von Hanseman cells) along with several small, round laminated Michaelis-Gutmann bodies (H&E high-power magnification).

Figure 4 Michaelis-Gutmann bodies are highlighted on a von Kossa stain (high-power magnification).

of oral antibiotics may be sufficient to achieve symptomatic and histological improvement.

Learning points

- ▶ Malakoplakia is a rare chronic inflammation that is characterised microscopically by Michaelis-Gutmann bodies, more commonly in immunocompromised patients.
- ▶ The presentation of malakoplakia is non-specific.
- ▶ Mainstay of treatment for malakoplakia is antibiotic therapy.

Contributor JMY, NWYS and GP conceptualized the study and wrote the article. FP provided valuable clinical images and description

Competing interests None declared.

Patient consent Obtained.

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