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Cronkhite-Canada syndrome with steroid dependency: A case report

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Author contributions: Jiang D collected basic patient information and imaging data and wrote the main content of the manuscript; Tang GD reviewed the literature and made the diagnosis; Lai MY and Huang ZN were responsible for the analysis and interpretation of data; Liang ZH was responsible for the conception and design of the study and the final approval of the version of the article to be published; All authors have read and approve the final manuscript.

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Abstract

BACKGROUND

Cronkhite-Canada syndrome (CCS) is a rare nonhereditary disease characterized by chronic diarrhoea, diffuse gastrointestinal polyposis and ectodermal manifestations. The lethality of CCS can be up to 50% if it is untreated or if treatment is delayed or inadequate. More than 35% of the patients do not achieve long-term clinical remission after corticosteroid administration, with relapse occurring during or after the cessation of glucocorticoid use. The optimal strategy of maintenance therapy of this disease is controversial.

CASE SUMMARY

A 47-year-old man presented to the hospital with a 3-mo history of frequent watery diarrhoea, accompanied by macular skin pigmentation that included the palms and soles, and onychodystrophy of the fingernails and toenails. Gastroscopy and colonoscopy revealed numerous polyps in the stomach and colon. After other possibilities were ruled out by a series of examinations, CCS was diagnosed and treated with prednisone. The patient took prednisone for more than 1 year before achieving complete resolution of his symptoms and endoscopic findings. The patient was then given prednisone 5 mg/d for 6 mo of maintenance therapy. With clinical improvement and polyp regression, prednisone was discontinued. Eight mo after the discontinuation of prednisone, the diarrhoea and gastrointestinal polyps relapsed. Therefore, the patient was given the same dose of prednisone, and complete remission was achieved again.

CONCLUSION

It is necessary to extend the duration of prednisone maintenance therapy for CCS. Prednisone is still effective when readministered after relapse. Surveillance endoscopy at intervals of 1 year or less is recommended to assess mucosal disease activity.

Key Words: Cronkhite-Canada syndrome; Endoscopy; Prednisone; Relapse; Maintenance therapy; Case report

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Core Tip: Cronkhite-Canada syndrome (CCS) is a rare gastrointestinal polyposis syndrome. Here, we report a case of CCS that has been followed for almost 4 years. The patient was treated with prednisone. After he discontinued prednisone, his clinical and endoscopic manifestations relapsed. The patient was given prednisone again, and it was effective in bringing about a second remission. It is necessary to extend the duration of prednisone maintenance therapy for CCS. Surveillance endoscopy at intervals of 1 year or less is recommended to assess mucosal disease activity.

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INTRODUCTION

Cronkhite-Canada syndrome (CCS) is a rare gastrointestinal polyposis syndrome characterized by dermatologic manifestations associated with chronic diarrhoea, malnutrition, and enteric protein wasting resulting from chronic inflammatory changes in the intestinal mucosa[1-3]. Since the disease was first described in 1955, only approximately 500 patients with CCS have been reported worldwide, among which over 70% were from Japan[2,4,5]. Although disease presentation has been well described, there is no consensus on the management of CCS. Its clinical course is characterized by progressive disease with occasional spontaneous remissions and frequent relapses[6]. The lethality of CCS can be up to 50% if it is untreated or if treatment is delayed or inadequate. The 5-year mortality of CCS patients can be as high as 55% secondary to complications[7].

CASE PRESENTATION

Chief complaints

A 47-year-old Chinese man presented to the hospital with frequent watery diarrhoea.

History of present illness

The patient had no history of prior illness and no family history of any similar disease.

History of past illness

The patient's symptoms had started 3 mo prior with frequent watery diarrhoea (4-5 times/d). For nearly a month, there was apparent blood in the diarrhoea and positive faecal occult blood with acid regurgitation, eructation, occasional nausea, and a weight loss of 10 kg within 2 mo. The patient was treated with symptomatic therapies, such as spasmolytics and antibiotics, which were ineffective in alleviating his symptoms.

Personal and family history

The patient had no history of prior illness and no family history of any similar disease.

Physical examination

A physical examination revealed marked alopecia; brownish macular pigmentation of the facial region, palms and soles; and onychodystrophy of the fingernails and toenails (Figure 1).

Laboratory evaluation

The laboratory findings included a positive faecal occult blood showing 0-2 red blood cells/haptoglobin, an albumin concentration of 32.7 g/L (normal range 40-55 g/L), and cytoplasmic antinuclear antibody positivity with a titre of 1:320.

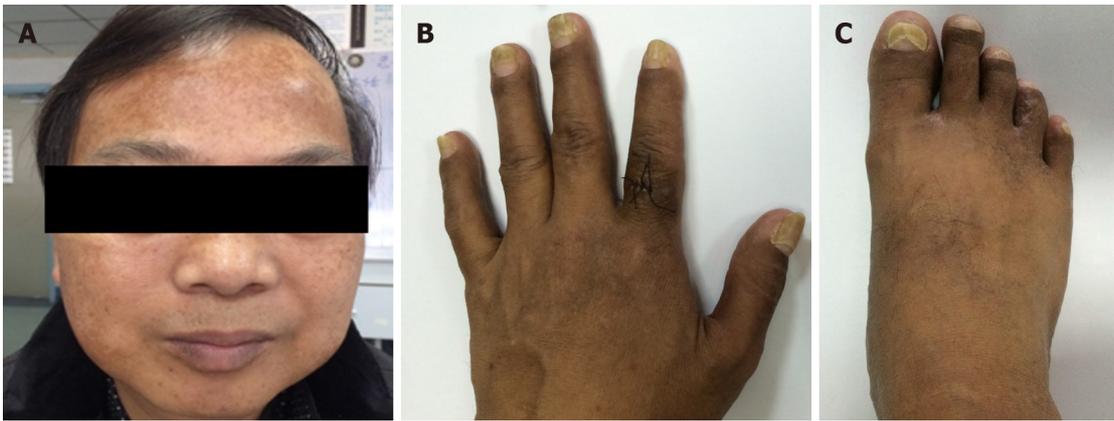


Figure 1 Physical examination findings included alopecia, brown macular pigmentation over the facial region, palms and soles, and onychodystrophy of the fingernails and toenails. A: Facial region; B: Palms; C: Soles.

Imaging evaluation

Gastroscopy revealed multiple polyps in the stomach and duodenum (Figure 2A). Endoscopic ultrasonography revealed diffuse thickening of the gastric mucosa to 8.5 mm (*i.e.* massive enlargement of the mucosa and submucosa), and the polyps originated from the mucosa, which was internally hyperechoic and inhomogeneous (Figure 2B). Colonoscopy revealed numerous polyps occupying the colonic and rectal mucosa (Figure 2C).

Further diagnostic work-up

Histological examination of biopsy specimens obtained from the colon and the stomach was consistent with hyperplastic polyps, and immunoglobulin G 4 staining was negative.

FINAL DIAGNOSIS

The final diagnosis of the present case was CCS.

TREATMENT

Oral administration of prednisone was initiated at 40 mg/d, and the daily steroid dose was varied in a tapered regimen, with a decrease of 5 mg every 4 wk.

OUTCOME AND FOLLOW-UP

After 3 mo of treatment, the gastrointestinal symptoms and ectodermal manifestations began disappearing one-by-one, and gastrointestinal endoscopy showed small, sparsely distributed polyps. After 6 mo of treatment, the patient's skin colour was normal and his nail dystrophy was improved (Figure 3A and B). Gastroscopy and colonoscopy showed that the polyps were significantly reduced in size and number. However, the patient's gastrointestinal manifestations relapsed, including diarrhoea; in response, we slowed the prednisone reduction, subtracting 5 mg from the daily dose only once every 3-6 mo. Until April 2018, the patient took only 5 mg of prednisone a day. After 14 mo of treatment, he was admitted to our hospital again for follow-up. The faecal occult blood test was negative; serum albumin levels had risen to normal. Additionally, the tumour markers were negative. Gastroscopy revealed several polyps with a diameter of 3-4 mm in the gastric antrum and duodenum. Some of the polyps were removed by endoscopic mucosal resection (Figure 3C). Colonoscopy revealed polyp regression (Figure 3D). Beginning in April 2018, the patient was given 5 mg of prednisone/d for 6 mo of maintenance therapy. With clinical improvement and polyp regression, prednisone was discontinued.

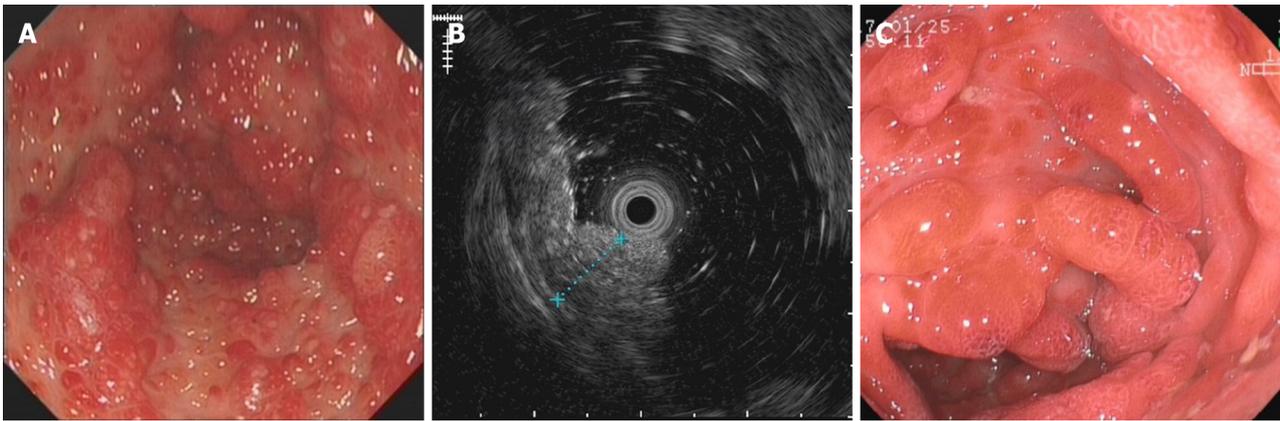


Figure 2 Gastroscopy. A: Gastroscopy revealed multiple polyps in the stomach; B: Endoscopic ultrasonography revealed diffuse thickening of the gastric mucosa of up to 8.5 mm in thickness; C: Colonoscopy revealed numerous polyps occupying the colonic and rectal mucosa.

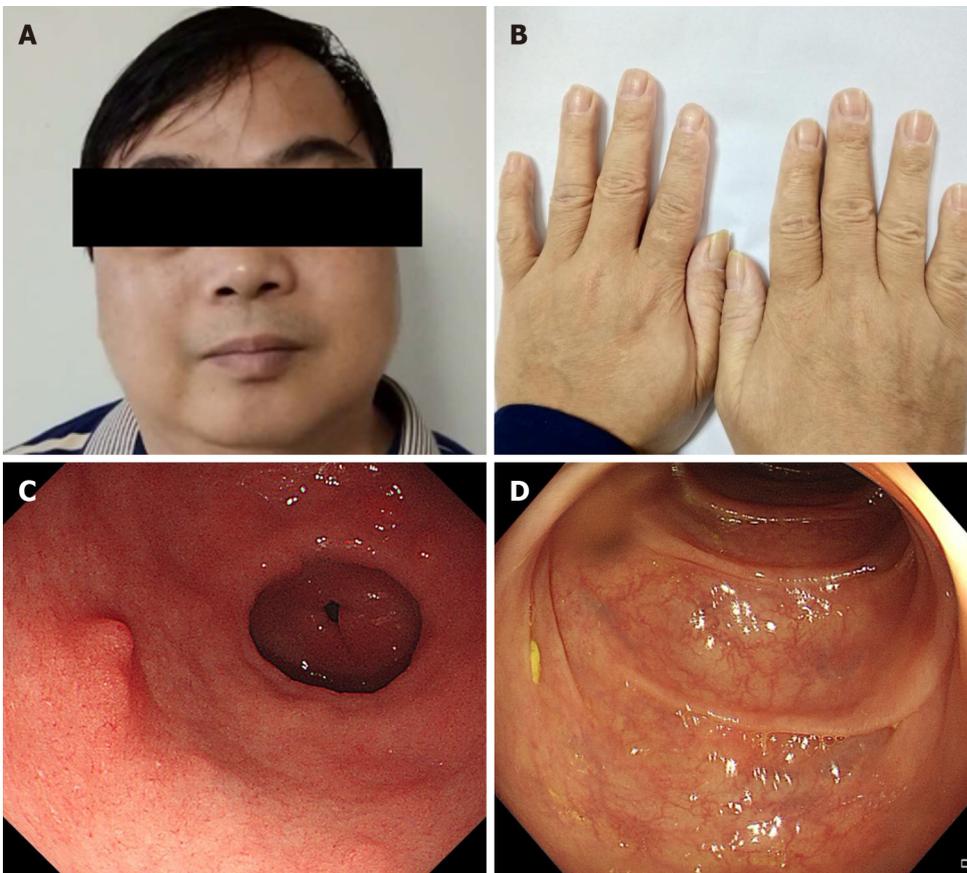


Figure 3 Examination after treatment. A: The patient showed great improvement in his skin pigmentation after 6-8 mo of steroid therapy; B: The patient showed great improvement in his nail dystrophy after 6-8 mo of steroid therapy; C and D: After 14 mo of treatment, gastroscopy revealed several polyps with a diameter of 3-4 mm in the gastric antrum (C), and colonoscopy revealed polyp regression (D).

Eight mo after the discontinuation of prednisone, the patient developed ectodermal manifestations again and was reviewed by gastroscopy, which suggested a relapse of CCS. We began to treat the patient again with the original regimen (*i.e.* a dose of 40 mg prednisone qd po with a decrease of 5 mg every 4 wk). Prednisone was reduced to 10 mg/d and maintained for 6 mo. In October 2020, we reviewed the patient by gastroscopy and colonoscopy, which indicated that the mucosal lesions had disappeared.

DISCUSSION

CCS is a rare disease, and there is no consensus on therapy at present. Treatment for CCS is largely based on anecdotal evidence and traditionally consists of nutritional support, histamine receptor antagonists, steroids, immunosuppression, or *Helicobacter pylori* eradication. A retrospective analysis has confirmed that steroid therapy is the mainstay of medical treatment and that 30-49 mg/d of orally administered prednisolone is optimal for active CCS, suggesting that the prednisolone dose should be slowly tapered only after endoscopic confirmation of the regression of polyposis[2]. The duration of therapy usually required is 6-12 mo. Once a sustained response is achieved, corticosteroids should be slowly tapered and eventually discontinued. Recurrences often respond to corticosteroid retreatment[8]. More than 35% of patients failed to achieve long-term clinical remission after corticosteroid administration, and relapse occurred during or after the cessation of glucocorticoid use. A proportion of patients were prescribed low-dose (5-10 mg/d) corticosteroids or immunosuppressants to counteract the tendency to relapse[5].

As immunological dysregulation is one of the important factors hypothesized to be present in CCS, the long-term use of immunoregulatory drugs or biologics may be useful for active or refractory disease[9]. Steroid-sparing therapies such as cyclosporine A and an antitumor necrosis factor- agent, a combination that has shown promise in a few cases, can be used in steroid-resistant cases to induce or maintain clinical remission. Other case reports described the beneficial response of immunosuppressive therapies including infliximab, sirolimus, tacrolimus, methicillin, and mycophenolate mofetil[2,10-14]. Azathioprine and mesalazine used with steroid maintenance therapy were reported to be associated with sustained clinical remission[3,15-17]. In a study by Mao *et al*[17], the patient was given a dose of 1.25 mg/kg/d azathioprine (previous reports had specified a dose of 2 mg/kg/d). The prednisone dose was tapered after 6 wk of therapy, and azathioprine was initiated. Corticosteroid side effects resolved when the prednisone was tapered, and CCS symptoms remained controlled on azathioprine without adverse effects; the total course of treatment consisted of 6 wk of prednisone and 26 wk of azathioprine. In a study by Schulte *et al*[3], discontinuation of steroid therapy was not possible, and mesalazine (1000 mg tid) was added to prednisolone (10.0 mg/d). The steroid dosage was further reduced over the course of 3 years; when all polyps had disappeared and the steroid therapy was finished, the dosage of mesalazine was reduced in a stepwise fashion. Four years later, the mesalazine was stopped, and more than 14.0 years after the initial diagnosis, the patient was still in complete remission without any treatment.

In conclusion, the mainstay of medical treatment for CCS is steroid therapy. Endoscopic remission and histopathologic remission are the therapeutic goals. A standard dose-adjustment protocol for prednisolone in CCS patients has not been established. Prednisone treatment after relapse in this case is still effective. Treatment should, therefore, be individualized for each patient according to their symptoms and recorded response to previous therapy. In episodically active clinical disease, the frequent use of steroids is necessary to prevent endoscopic relapse. Azathioprine, mesalazine, and other drugs merit consideration as CCS maintenance therapy. Surveillance endoscopy at intervals of 1 year or less is recommended to assess mucosal disease activity.

CONCLUSION

Steroid therapy is the mainstay of medical treatment for CCS. Endoscopic remission and histopathologic remission are the therapeutic goals. In the current case, prednisone treatment was still effective after relapse. Treatment should, therefore, be individualized for each patient according to his or her symptoms and recorded responses to previous therapy. Surveillance endoscopy at intervals of 1 year or less is recommended to assess mucosal disease activity.

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