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Plummer-Vinson Syndrome Associated With Crohn's Disease: A Case Report

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SUMMARY
Plummer-Vinson syndrome (PVS) is a rare condition with the classic triad of dysphagia, anaemia and oesophageal ring. Crohn's disease is generally accompanied by malnutrition and iron deficiency. Two cases of patients with Crohn's disease presenting with PVS have been described in the literature. We present a case of PVS complicating Crohn's disease in a 22-year-old female patient with progressively worsening dysphagia.

Key words: *Plummer Vinson syndrome - Crohn's disease*

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I. Introduction

Plummer-Vinson (or Paterson-Kelly) syndrome is a rare condition, presenting as a classic triad of dysphagia, iron deficiency anaemia and oesophageal ring. The association of this syndrome with Crohn's disease is uncommon. We report a case of VPS associated with Crohn's disease.

II. Clinical case

Patient aged 22, chronic smoker of 03 packets per year, chronically weaned off cannabism, occasional alcoholic; bipolar disorder under treatment (olanzapine, sulpiride and maprotiline hydrochloride) and mother treated for UC (deceased); the history of the disease dates back to the age of 10, the diagnosis of colonic Crohn's disease classified as A2L2B1 was retained on clinical, morphological and histological criteria, with a diagnostic delay of 9 years. She was admitted in January 2018 with bloody diarrhoea with 3 to 6 stools per day, Kœnig's syndrome, clinical anaemia and functional dysphagia. On clinical examination, the patient was in good general condition (WHO 0) with a BMI of 20 kg/m2 and discoloured conjunctivae.

Biologically, the patient presented with anaemia of 8.6 g/dl, microcytic hypochromia, mild hyperleukocytosis of 10380 E/mm3, thrombocytosis of 799000 E/mm3, high CRP of 30 mg/l, correct albumin of 37 g/L, and hypoferritinaemia of 3 mg/ml.

Entero-CT showed no thickening of the cecum, respect for the ICD, diffuse, moderate circumferential colonic parietal thickening measuring 7 mm in thickness, and no deep tract or collection.

A short colonoscopy revealed multiple deep and superficial ulcerations with no spaces of healthy mucosa. Histological examination showed subacute ulcerative rectitis, with no glandular differentiation.

A FOGD was performed, revealing a circumferential ring in the upper 1/3 of the oesophagus, dilated by endoscope without immediate incident. Multiple biopsies were performed to rule out other causes of hypoferritinaemia, and showed no HP, no celiac disease (LEL 17% without atrophy) and no histological signs in favour of Biermer's disease. And given that the patient is young and of childbearing age (with an irregular menstrual cycle), a pelvic ultrasound was performed to rule out gynaecological causes, which came back without any particularity.

The diagnosis of Plummer Vinson syndrome due to hypoferritinemia in luminal colonic Crohn's disease classified as A2L2B1 active was accepted; the patient was put on corticosteroid therapy for her Crohn's disease, and received 04 sessions of iron infusion; with a good clinical response (disappearance of diarrhoea, abdominal pain and anaemic syndrome); but the patient was lost to follow-up for two years. The patient consulted for a relapse of her Crohn's disease, associated with high dysphagia of functional appearance (old, intermittent, paradoxically capricious); a fibroscopy had shown a recurrence of her impassable oesophageal ring. The patient underwent endoscopic dilatation with Savary-Gilliard candles to a diameter of 12 mm, and was put on a martial treatment (iron infusion) with therapeutic education.

The patient was lost to follow-up for 2 years (non-disciplined patient), and was then admitted to our department for a severe clinical and endoscopic relapse of her disease (CDEIS 25); associated with high dysphagia of functional appearance; a FOGD made objective of a circumferential ring at the level of the upper 1/3 of the

oesophagus, dilated by Savary-Gilliard candles to a diameter of 12 mm without immediate incident; as well as a duodenal location of Crohn's disease.

The patient was put on injectable corticosteroids for 03 days, followed by oral corticosteroids, iron infusion and specialist psychiatric care. The patient is a candidate for anti-TNF treatment.

III. Discussion

The precise pathogenesis of PVS is still unknown, although iron deficiency has been reported as an acceptable risk factor, as suggested by the positive impact of iron supplementation on symptom improvement. However, malnutrition, genetic predisposition or autoimmune conditions such as rheumatoid arthritis, celiac disease or thyroid disease may also play an important role [1].

Crohn's disease has been described to be associated with malnutrition and iron deficiency, which is the most common extra-intestinal disorder [5]. Specifically, female gender, disease activity, previous surgery and use of immunomodulators or biologics are considered risk factors for iron deficiency in Crohn's disease [6]; our patient fulfilled two of these factors.

Progressively worsening dysphagia is the patients' main complaint. Endoscopy, to exclude obstructive lesions, reveals the annulus as a smooth grey mucosal projection that reduces the luminal diameter, and upper gastrointestinal tract exploration is necessary to exclude common PVS-related conditions such as celiac disease [3].

Iron supplementation is indicated in all cases to relieve dysphagia, but the ring does not resolve, requiring mechanical dilatation using a balloon or Savary-Gilliard candles, which relieve symptoms in almost 94% of cases [4].

Iron deficiency is the most common systemic complication associated with inflammatory bowel disease (IBD). Crohn's disease can lead to severe anaemia due to dietary deficiencies, haemorrhage, intestinal iron malabsorption, inflammation and adverse drug reactions [2].

Our patient suffered from iron deficiency due to active Crohn's disease. Another case of Crohn's disease and PVS highlighted poor compliance with IBD treatment as a risk factor [7]. Finally, endoscopic dilatation and iron supplementation is necessary; adequate treatment and control of IBD activity appear to be important for the prevention of PVS.

PVS has been associated with cancer of the mouth, hypopharynx and stomach, and with squamous cell carcinoma of the oesophagus in 3 to 16% of cases [1], so endoscopic surveillance is advisable.

IV. Conclusion

The association between Crohn's disease and Plummer-Vinson syndrome remains uncommon. A high index of suspicion and a clinical perspective are necessary for early diagnosis of PVS and its complications. Given the risk of PVS degeneration, endoscopic surveillance is recommended.

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