Gavric et al., J Clin Case Rep 2017, 7:8
DOI: 10.4172/2165-7920.10001014

Journal of Clinical Case Reports

Case Report OMICS International

Sarcoidosis and Collagenous Colitis – Important Clinical Association or Coincidence?

Aleksandar Gavrić*, David Drobne, Nina Zidar and Borut Štabuc

University Medical Center Ljubljana, Ljubljana, Slovenia

Abstract

We present a case of a 57-year-old woman with two rare concomitant diseases; sarcoidosis and collagenous colitis. Patient was admitted to our hospital with the symptoms of watery diarrhea that intermittently lasted for years because of delayed diagnosis. Despite increasing awareness of microscopic colitis, the delayed diagnosis remains an important problem. Diagnosis was quickly confirmed with flexible proctosigmoidoscopy. Rectal biopsies were sufficient for diagnosis. Symptoms improved dramatically the second day of the induction therapy with budesonide. Causal relationship between sarcoidosis and microscopic colitis is not yet confirmed, and to our knowledge, this is the first such case report.

Keywords: Chronic watery diarrhea; Collagenous colitis; Calprotectin; Budesonide; Sarcoidosis

Introduction

Collagenous colitis (CG) was first described in 1976. It classically occurs in female patients with a peak incidence around 60 to 70 years [1]. CG may be associated with celiac disease and the use of certain medications, particularly nonsteroidal anti-inflammatory drugs and proton pump inhibitors. There are also reports that CG may be precipitated by infections [2]. Sarcoidosis is an inflammatory disease with unknown etiology. The cardinal feature are noncaseating granulomas. The incidence of intestinal involvement is not known as there are few reported cases in literature. Some authors estimate it to less than 1%. Stomach is most commonly affected, colon less frequently [3]. Causal relationship between sarcoidosis and CG has not been confirmed.

Case Report

A 57-year-old Caucasian woman was admitted to our gastroenterology department with persistent watery diarrhea since 2010. Diarrhea was cyclic, and the last episode was most severe with ten bowel movements per day and night. She complained of fatigue, weight loss, crampy lower abdominal pain and weight loss of five kilograms in last months. Week before admission her calprotectin was checked in an outpatient clinic and was increased (>500 mcg/g). Coprocultures were negative. Blood work was normal and physical examination did not reveal abnormalities which was the reason she was initially planned for outpatient diagnostics. Her medical history was notable for hypothyroidism, migraines, and sarcoidosis of the nose and lymph nodes, which was confirmed by the biopsy from the nasal skin. She was regularly seen by pulmonologist who suggested corticosteroid therapy, but she rejected therapy. She underwent esophagogastroduodenoscopy in 2011, celiac disease and *Helicobacter pylori* infection were ruled out. She was seen by proctologist in 2014 and had colonoscopy without biopsies as colon mucosa was normal. She was taking sodium levothyroxine, pregabalin, and sumatriptan. Her family history was unremarkable. She worked as a cook woman. She did not smoke or drink alcohol.

On the first day of admission, we performed flexible proctosigmoidoscopy with rectal biopsies. Rectal mucosa was hyperemic and fragile with touched bleeding. Histopathological exam revealed intraepithelial lymphocytes (15 per 100 epithelial cells) and granulocytes. Trichrome staining revealed 20 μm thick collagen band under the basal membrane, findings suggestive for CG. We started induction therapy with budesonide 9 mg per day and patient experienced dramatic çlinical improvement. All other tests were cancelled and patient was discharged home. Patient was seen two months after the end of the eight-week therapy with budesonide. Her symptoms deteriorated one month after she finished therapy. Because of very high calprotectin concentration and the history of the bloody stools colonoscopy was later performed to exclude concomitant inflammatory bowel disease. Colon mucosa appeared endoscopically normal. Pathological exam of colon biopsies again revealed collagenous microcolitis. Upper endoscopy was performed again and was normal. Biopsies of the duodenum revealed increased lymphocytic epithelial infiltration. The maintenance therapy with the Budesonide 9 mg was reinduced and patient scheduled for a follow-up (Figures 1 and 2).

Discussion

Sarcoidosis is a multisystemic disorder of unknown etiology and has the potential to affect almost every tissue in the body [4]. Gastrointestinal involvement is very rare (less than 1%). The stomach is most commonly affected and the colon least frequently [5]. The sarcoidosis of the colon is usually asymptomatic [6] but when symptoms occur the clinical

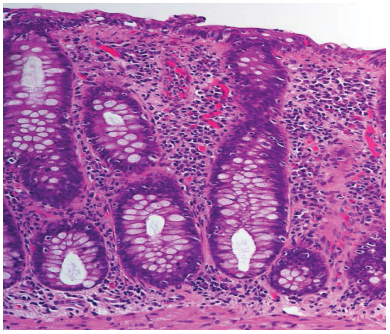

Figure 1: Collagenous colitis. Mild inflammation of the lamina propria, preserved crypt architecture and subepithelial collagen band.

***Corresponding author:** Aleksandar Gavrić, University Medical Center Ljubljana, Ljubljana, Slovenia, Tel: +38641948332; E-mail: aleksandar.gavri@gmail.com

Received July 05, 2017; **Accepted** August 23, 2017; **Published** August 27, 2017

Citation: Gavrić A, Drobne D, Zidar N, Štabuc B (2017) Sarcoidosis and Collagenous Colitis– Important Clinical Association or Coincidence? J Clin Case Rep 7: 1014. doi: 10.4172/2165-7920.10001014

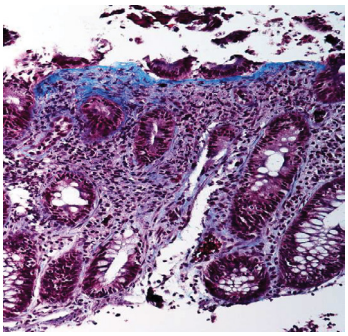

Figure 2: Trichrome staining with blue staining of the collagen band.

presentation of gastrointestinal sarcoidosis commonly resemble those of an obstructive colonic disease [5]. A case of an isolated gastrointestinal sarcoidosis with the diarrhea being the chief complaint was described [6,7]. Biopsies in this case revealed histological evidence of sarcoidosis and patient improved after the therapy with methylprednisolone, whether in our case, biopsies of the colon and upper GI tract did not show changes specific for sarcoidosis but for CG. The common point with our case is delayed diagnosis. Both patient presented with chronic watery diarrhea and had prior colonoscopies without biopsies as mucosa was normal. Macroscopic lesions in colon are seen only in 25% cases of gastrointestinal sarcoidosis, which warrant low treshold for biopsies despite normal colon mucosa [6]. The main difference is that our patient had a concomitant diagnosis of CG causing diarrhea and was thus diagnosed with two rare diseases; sarcoidosis and CG. Sarcoidosis itself was not explained as the etiology of diarrhea. The coincidence of both diagnoses captured our attention as such case has not been published yet.

Before performing total colonoscopy, we performed flexible proctosigmoideoscopy with biopsies to exclude ulcerative colitis. Pathohistological exam of rectal mucosa showed changes typical for CG. Precise history revealed that patient experienced episodes of spontaneous resolutions of watery diarrhea six years before admission. And indeed, recent long-term studies have suggested that CG usually runs a benign clinical course where in some cases symptoms resolve with no treatment or remission occurs with minimal therapy. Gastrointestinal symptoms have been reducing quality of life in this patients for at least six years which were dramatically improved after the therapy with budesonide. Recent prospective microscopic colitis registry from the USA revealed that the average duration from time of diarrheal symptoms to microscopic colitis diagnosis was 2.5 years [8].

Some authors suggest to biopsy all segments of colon when looking for microscopic colitis [9], because mucosa can have patchy pattern of histological changes and because changes are more frequently presented in the right rather than in left colon. The fact that typical changes were seen in the rectal mucosa can be explained by the high inflammatory burden and long-lasting disease and diagnosis in such cases diagnosis can be achieved by flexible proctosigmoidoscopy. High calprotectin measured in our patient shows that this marker of luminal colon inflammation can be increased in CG despite endoscopically normal mucosa. CG should be differential diagnosis when calprotectin levels are high. We think that endoscopy in patients with sarcoidosis presented with chronic watery diarrhea must not be postponed. Biopsies of the colon mucosa must be obtained to exclude CG and colonic sarcoidosis, which are both possible etiologies of chronic watery diarrhea.

Conclusion

This case may indicate the higher incidence of the CG in patients with sarcoidosis. We are aware that coincidence of these two diagnosis in our patient can also be accidental. It is known that many patients with IBD have concomitant sarcoidosis. Recent study by Fischer et al. [10] revealed common susceptibility genes that are associated with IDB and sarcoidosis. Similair study with the comparison for genetic background in CG patients has not yet been reported. Cases as ours could foster such further studies.

References

1. Toole A (2016) Optimal management of collagenous colitis: A review. Clin Exp Gastroenterol 9: 31-39.
2. Freeman HJ (2012) Long-term natural history and complications of collagenous colitis. Can J Gastroenterol J Can Gastroenterol 26: 627-630.
3. Daldoul S, Triki W, El-Jeri K, Zaouche A (2012) Unusual presentation of a colonic sarcoidosis. Case Rep Med 2012: 169760.
4. Storch I, Sachar D, Katz S (2003) Pulmonary manifestations of inflammatory bowel disease. Inflamm Bowel Dis 9: 104-115.
5. Erra P, Crusco S, Nugnes L, Pollio AM, Di Pilla G, et al. (2015) Colonic sarcoidosis: Unusual onset of a systemic disease. World J Gastroenterol 21: 3380-3387
6. Ghrenassia E, Mekinian A, Chapelon-Albric C, Levy P, Cosnes J, et al (2016) Digestive-tract sarcoidosis: French nationwide case-control study of 25 cases. Medicine (Baltimore) 95: e4279.
7. Stemboroski L, Gaye B, Makary R, Monteiro C, Eid E (2016) Isolated gastrointestinal sarcoidosis involving multiple gastrointestinal sites presenting as chronic diarrhea. ACG Case Rep J 21: 3.
8. Yen EF, Yoo J, Bianchi LK, Moller NA, Goldberg MJ. (2016) Sa1917 Delayed Diagnosis in Microscopic Colitis: Analysis of a Large Prospective Cohort Gastroenterology 150: S403.
9. Hilpüsch F, Johnsen PH, Goll R, Valle PC, Sørbye SW, et al. (2016) Microscopic colitis: A missed diagnosis among patients with moderate to severe irritable bowel syndrome. Scand J Gastroenterol 31: 1-5.
10. Fischer A, Nothnagel M, Franke A, Jacobs G, Saadati HR, et al. (2011) Association of inflammatory bowel disease risk loci with sarcoidosis, and its acute and chronic subphenotypes. Eur Respir J 37: 610-616.

Citation: Gavrić A, Drobne D, Zidar N, Štabuc B (2017) Sarcoidosis and Collagenous Colitis– Important Clinical Association or Coincidence? J Clin Case Rep 7: 1014. doi: 10.4172/2165-7920.10001014