



SHORT REPORT

“Ulcerative crepitus” — A case with subcutaneous emphysema and pneumomediastinum without colonic perforation or toxic megacolon in ulcerative colitis successfully treated conservatively

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KEYWORDS

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Abstract

A 19-year-old man with a 1-year history of ulcerative colitis presented with fever, bloody diarrhea and severe dehydration. He was on *po*.48 mg methylprednisolon and 3 g mesalazine daily, and has recently finished taking chlarythromycin for *Campylobacter jejuni* infection. On physical examination, no abdominal tenderness was found, but surprisingly, extensive bilateral subcutaneous emphysema was detected in the supraclavicular regions. Laboratory tests proved anaemia, elevated white blood cell count, thrombocyte count and CRP levels. Stool culture was negative. Chest X-ray and CT scan revealed pneumomediastinum and subcutaneous air on the neck spreading to the scapular regions. Besides blood transfusion, *iv.* cyclosporin therapy was initiated (200 mg/day) along with *iv.* methylprednisolon (1 mg/kg/day) and *iv.* ceftriaxon (2 g/day). Stool frequency and bloody stools decreased remarkably within one week, and subcutaneous emphysema has resolved. Colonoscopy one week later revealed deep, extensive ulcerations in the transverse and descending colon without any sign of previous perforation. Cyclosporin and methylprednisolon was continued orally.

Pneumomediastinum and subcutaneous emphysema in ulcerative colitis are unusual complications, typically linked to retroperitoneal colonic perforation or toxic megacolon, and are extremely rare without preceding endoscopic procedures. Except from two cases in the literature, conservative treatment with *iv.* antibiotics and steroids failed to save from urgent surgical

Abbreviations: *C. jejuni*, *Campylobacter jejuni*; UC, ulcerative colitis.

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procedure, resulting in a partial or total colectomy. In our case we were able to avoid urgent surgery by the immediate use of *iv.* cyclosporin in combination with *iv.* steroids and antibiotics, while the outcome of the bowel remains questionable in the next few months.

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1. Introduction

Pneumomediastinum and subcutaneous emphysema in ulcerative colitis (UC) are unusual complications, which are extremely rare without preceding endoscopic procedures.^{1,2} These phenomena are typically linked to retroperitoneal colonic perforation following sigmoido- or colonoscopy^{3,4} or to a markedly dilated toxic megacolon,^{1,5} and treatment often requires urgent surgical intervention. We report a case of a severe UC where mediastinal and subcutaneous air appeared without previous endoscopic examinations, and which could be managed conservatively by the use of *iv.* cyclosporin in combination with *iv.* steroids and antibiotics.

2. Case report

A 19-year-old man with a 1-year history of UC presented in our hospital with fever, bloody diarrhea and severe dehydration. Following a previous relapse he was on a daily 48 mg methylprednisolone and 3 g mesalazine therapy *po.*, and has recently finished taking chlarythromycin prescribed for an infection of *Campylobacter jejuni* (*C. jejuni*). On physical examination, pale skin and thrush were noted, no abdominal tenderness was found. Surprisingly, extensive bilateral subcutaneous emphysema was detected in the supraclavicular regions, on the shoulders and the upper back, which, according to the patient, had developed 1 week earlier accompanied by a short period of upper chest and neck pain, and a slight shortness of breath. Laboratory tests proved anaemia (haemoglobin: 68 g/L), elevated white blood cell count (16.83 G/L), thrombocyte count (460 G/L) and CRP levels (32.4 mg/L). Stool culture on admission showed no infection of *C. jejuni*. Chest X-ray and CT scan revealed a 10–15 mm wide pneumomediastinum and subcutaneous air trapped on the neck spreading to the scapular regions, without the presence of a pneumothorax (Fig. 1). The CT scan also showed some free air infiltrating both crus of the diaphragm, which could be followed in the retroperitoneum by an abdominal X-ray, while no intraperitoneal air or colonic dilatation was present. No intra-abdominal free fluid was shown by the abdominal ultrasound, which would suggest peritonitis related to colonic perforation. The patient has received blood transfusion and *iv.* cyclosporin therapy was initiated (200 mg/day) along with *iv.* methylprednisolone (1 mg/kg/day) and *iv.* ceftriaxone (2 g/day). The patient quickly responded to therapy, stool frequency and bloody stools decreased remarkably within one week, and subcutaneous emphysema has resolved, confirmed by a chest X-ray. Colonoscopy was performed one week later to evaluate the degree of inflammation, which revealed deep, extensive ulcerations in the transverse and descending colon without any sign of previous perforation (Fig. 2). Cyclosporin and methylprednisolone therapy was continued orally.

3. Discussion

Pneumomediastinum and subcutaneous emphysema without preceding trauma can be produced by gas leaking along fascial tissues even from distant origins, such as the bowel.⁶ Symptoms include severe chest and neck pain, dyspnea and changes in voice, while physical signs consist of subcutaneous crepitation on the neck and Hamman's sign, a crackling sound on auscultation congruent with the cardiac cycle.^{7,8} Small bilateral pneumothoraces may also appear as a consequence of a rupture of the mediastinal parietal pleura, secondary to the increased mediastinal pressure.^{1,5} Beyond physical examination, diagnosis is based on chest and abdominal radiography and preferably a CT scan. In most UC cases complicated with pneumomediastinum, free intraperitoneal air could not be detected,^{1,2,5,9,10} while retroperitoneal air was shown in nearly half of them.^{2,3,9} Colonic perforation in UC is often associated with toxic megacolon, nevertheless, it can appear also without dilatation, while preceding endoscopic procedures or barium enema may increase its risk significantly. Retroperitoneal air does not necessarily mean an actual perforation, as extensive, deep ulcerations may permit gas leakage without the presence of a detectable discontinuity of the colonic wall,^{1,5} similarly to our case. At the time of the colonoscopy, the pneumomediastinum and subcutaneous emphysema have been disappeared, and no further subcutaneous air developed, suggesting the absence of a colonic



Figure 1 CT scan shows pneumomediastinum and subcutaneous emphysema (arrows) at the time of admission.

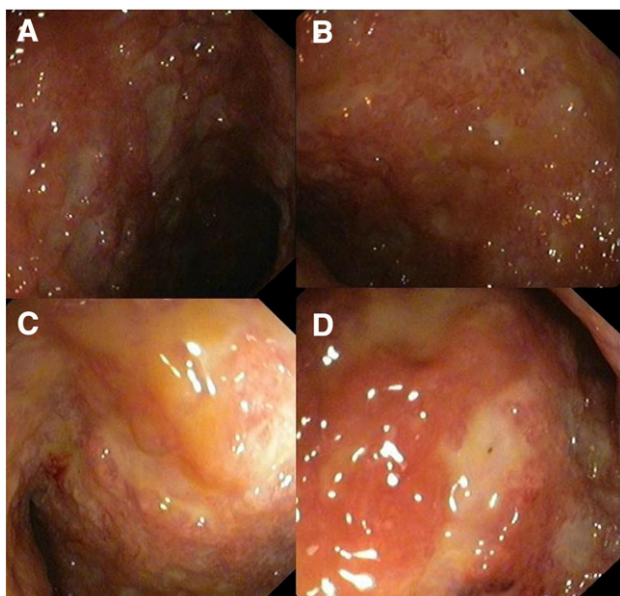


Figure 2 Colonoscopy performed one week later reveals deep, extensive ulceration in the transverse and descending colon.

perforation. Still, we have performed the colonoscopy one week after the admission of the patient, therefore a previous small perforation, already resolved by the time of examination, cannot be excluded. *C. jejuni* has been suggested to contribute to the pathogenesis of IBD in susceptible individuals,¹¹ however, a link between *C. jejuni* infection and pneumomediastinum has never been established. Stool culture has shown that *C. jejuni* infection was already resolved at the time of admission, nevertheless, this pathogen may have participated in the aggravation of colitis and thus in the development of severe complications. Due to the low number of cases, there are no established therapeutic protocols for relapses in UC complicated with pneumomediastinum. In general, conservative treatment is initiated under close observation, and immediate surgical intervention is employed in case of the signs of peritonitis or in the lack of clinical improvement within a few days. However, except from two cases available in the international literature,^{2,9} conservative treatment with parenteral nutrition, iv. antibiotics and steroids failed to save from urgent surgical procedure, resulting in a partial or total colectomy.^{1,3,5,10} In our case we were able to avoid urgent surgery by the immediate use of iv. cyclosporin combined

with iv. steroids and antibiotics, while the outcome of the bowel remains questionable in the next few months.

Conflict of interest

No conflict of interest exists.

Acknowledgements

AA was physician in charge of the patient, and drafted the manuscript. PI was the radiologist performing and evaluating the diagnostic imaging of the patient. NF, WT and MT were gastroenterologists treating the patient, they have corrected and improved the manuscript. All authors read and approved the final manuscript.

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