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CASE REPORT

Subcutaneous emphysema: a rare manifestation of a perforated diverticulitis in a patent inguinal canal

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Abstract Patients with complicated diverticulitis rarely present with extraperitoneal manifestations but the manifestation of subcutaneous emphysema appears even more seldom. We present the case of a patient with a history of diabetes and immunosuppression, who was admitted with sepsis in association with cellulitis and subcutaneous emphysema of the left groin. The absence of peritonism due to corticosteroid treatment, a history of a recent fall with an ilio- and ischiopubic fracture and subcutaneous emphysema led to a delay in the diagnosis. The final diagnosis was a perforated diverticulitis in a patent inguinal canal, which was only revealed after surgery. The various complications of diverticulitis, including extraperitoneal manifestations, and associated microorganisms implicated in cellulitis and subcutaneous emphysema are briefly reviewed.

Keywords Diverticulitis · Inguinal canal · Subcutaneous emphysema

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Introduction

One-third of patients with diverticulosis will develop diverticulitis. Intra-abdominal perforation, abscess, hemorrhage, intestinal stricture or obstruction and fistula are the most common complications. These patients frequently present with abdominal symptoms and, on occasion, with peritonism. However, in rare cases, complications outside of the peritoneal cavity have been reported [1].

Case report

An 82-year-old institutionalized female patient with a history of corticosteroid (2 × 40 mg/day) treatment for rheumatoid arthritis was admitted to our emergency department for dyspnea and left groin pain. These symptoms had appeared a week before admission, following a fall onto the left hip. Her medical history included corticosteroid-induced diabetes, arterial hypertension, ischemic cardiopathy and peritonitis.

Physical examination revealed a patient in a markedly diminished general condition, afebrile, tachypneic, tachycardic and hypotensive. The abdominal examination showed a previous laparotomy scar and predominant pain in the left lower quadrant on palpation, without signs of peritonism. A digital rectal examination could not be carried out due to pain.

The left groin showed an inflammatory swelling with crepitations upon palpation. The surrounding skin was ischemic with a beginning of central necrosis. Blood tests confirmed an inflammatory syndrome with high non-segmented neutrophiles and high C-reactive protein in addition to a discrete rhabdomyolysis, diabetes

decompensation and acute renal failure. A pelvic X-ray showed subcutaneous air and a left ilio- and ischio-pubic fracture (Fig. 1).

Systemic inflammation response syndrome (SIRS) along with the physical examination and pelvic X-ray described above led to the following differential diagnoses: rupture of a hollow pelvic organ consecutive to pelvic fracture, cutaneous infection in relation to the fall (with no evidence of portal of entry) or an incarcerated inguinal hernia with secondary rupture.

In addition to initiating resuscitation and antibiotic treatment, we performed an unenhanced – due to acute renal failure – abdominal-pelvic computed tomography scan (CT) with contrast media in the bladder to investigate the possibility of a fistula or a ruptured bladder. The CT scan showed thickened intestinal loops in the lower left quadrant with a suspicion of communication with the subcutaneous air

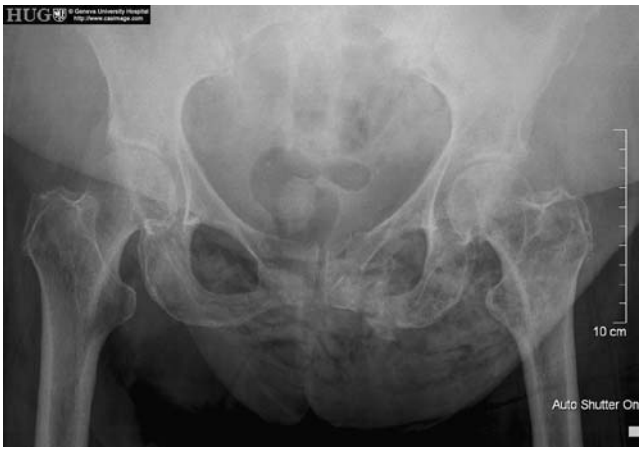

Fig. 1 Pelvic X-ray of the 82-year-old female patient showing subcutaneous air and a left ilio- and ischio-pubic fracture

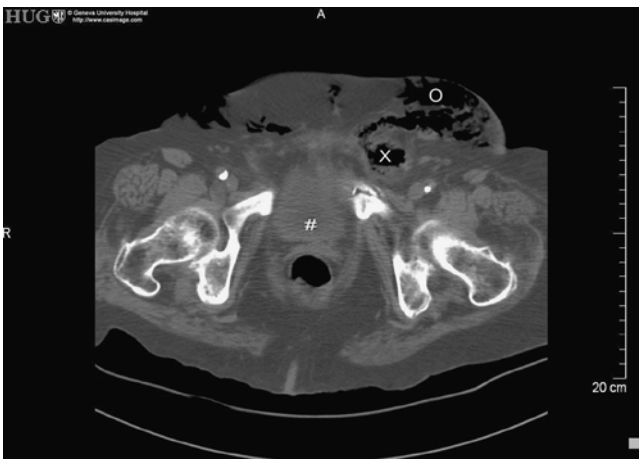

Fig. 2 Computed tomography scan of abdomen and pelvis of the 82-year-old patient. # Foley catheter, X air containing collection, O subcutaneous emphysema

associated with infiltration of the surrounding fat (Fig. 2). Radiological findings were consistent with an old left minimally displaced ilio- and ischio-pubic fractures due to the presence of a callus. There was neither a bladder breach nor an intestinal lesion; there was also no free air or free fluid in the peritoneal cavity.

An emergency surgical exploration of the groin region was decided upon. This showed cutaneous necrosis with an underlying abscess in an opening (containing no intestinal loop) leading to the peritoneal cavity. That opening was lateral, above the inguinal ligament through the direct floor. The laparotomy revealed a sigmoiditis, perforated along a length of 1 cm, with an important inflammatory adhesion against the patent inguinal canal, which was observed from the outside; there was no other extension of the diverticulitis inside the peritoneal cavity. An Hartmann's sigmoidal resection with terminal colostomie was carried out. The hernia defect was closed with five separated stitches of 2.0 thread. Skin and tissue necrosis was extensively resected, the skin was closed with four loose separated stitches and a drain was left in place.

The surgical resection specimen confirmed diverticulosis associated with a perforated diverticular abscess. The final diagnosis was a perforated diverticulitis in a patent inguinal canal leading to subcutaneous abscedation and gas gangrene. In the postoperative period, our patient developed a progressive necrosis of the abdominal wall with septic shock and multiple organ failure which unfortunately led to her death.

Discussion

To our knowledge, this particular type of presentation of a perforated diverticulitis has never been described. The most common intra-peritoneal complications of diverticulitis are abscess formation (46%), frank perforation (21%), diverticular hemorrhage (13%), intestinal stricture and occlusion (10%) and fistula (10%) [2]. Among the latter, colo-vesical fistulas are the most frequently encountered, followed by colo-vaginal, colo-uterine and finally colo-cutaneous fistula (1–2% of all fistula cases) in decreasing order of prevalence.

Fistulas occur in more than 90% of cases after resection surgery for acute diverticulitis [3] or after percutaneous catheter drainage of an intra-abdominal abscess. Only a minority of the fistulas occur spontaneously, without any abdominal pathology, with corticosteroid treatment being a risk factor [4]. However extraperitoneal manifestations of diverticulitis are rare and result from fistulizations outside of the peritoneal cavity. Most reported cases are retroperitoneal fistulas

presenting as thigh cellulites [5] that can also extend to the mediastinum and present thoracic or cervical subcutaneous emphysema [6]. Very few cases have been reported of a communication with mesenteric or portal veins involving the liver [7], the area of the appendix or epidural space. Subcutaneous emphysema can also result from a remote infection, and some dermatological manifestations are also described in the case of diverticulitis [8]. Our patient, who was on corticosteroid treatment for rheumatic arthritis [9], presented a clinically silent diverticulitis with this rare extra-peritoneal manifestation. The lower resistance of the patent's inguinal canal possibly allowed a perforation of the diverticular inflammation at this location.

Following pelvic trauma, one should suspect cutaneous infection secondary to local bacterial contamination. Once these microorganisms, typically *Clostridium perfringens*, have passed the skin barrier and are in anaerobic conditions [10], they can develop and produce toxins. The incubation period ranges from several hours to a few days. Cellulitis or necrosis then progresses to form an abscess, which ultimately develops into a severe systemic disease. Apart from *Clostridium perfringens*, *Streptococcus* group A or group B and *Staphylococcus* are frequently found in soft tissue infections, the former commonly encountered in diabetic patients. However, *Enterobacteria*, *Bacteroides fragilis* and *Enterococcus*, which are most commonly found in the peritoneal cavity, can also be involved. Nevertheless, gas gangrene essentially occurs as a result of infection by *Clostridium* species or other fermentation bacteria able to produce gas. No cutaneous portal of entry was found in our patient. The groin abscess was positive for *Bacteroides fragilis* and *Clostridium perfringens*, and the blood cultures were positive for *Escherichia coli* and *Enterobacter*, all of which are intestinal microorganisms.

Conclusion

Extra-abdominal rupture or fistulas are uncommon and atypical presentations of diverticulitis. Our patient showed very few specific symptoms in relation to the important immunosuppression by corticosteroids, making the diagnosis all the more difficult.

Immunosuppressed patients presenting with minimal digestive symptoms should receivecomplete radiological examinations to exclude the most probable diagnoses and extend potential diagnoses to the more unexpected possibilities. However, in the present case, the final diagnosis was only revealed after surgery.

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