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Case Report

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Acute Onset of a Swollen Leg With Crepitus: A Complication With Rectal Perforation

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Crepitus happened into the soft tissue is indicated subcutaneous emphysema. Subcutaneous emphysema of leg without trauma was likely to diagnose gas gangrene or gas-forming myonecrosis (GFM) at emergency department (ED). On the other hand, abdominal fatal condition with gas from the gut may spread to the leg should be considered a different diagnosis. We report a case of critically ill patient who presented to the ED with initial features suggestive of necrotizing fasciitis with gas gangrene of left leg. Assessment and further intervention revealed subcutaneous emphysema of leg secondary to a perforation of rectum associated with previous anastomosis site for rectal cancer surgery. Subcutaneous emphysema of the leg rarely happened secondary to perforation of the gastrointestinal tract and has often created serious diagnostic problems which may lead to mortality. Consequently, prompt diagnosis and aggressive treatment is imperative. Physicians and surgeons should be aware of this condition that could be fatal but curable by early intervention.

Key words: *subcutaneous emphysema, rectal perforation, gas gangrene*

Introduction

Gas in the extremities often prompts emergency department (ED) physicians to diagnose gas-forming myonecrosis (GFM) or gas gangrene. GFM or gas gangrene in the extremities was a fatal bacterial infection would make the ED physicians only focus on the involved area. However, other sources of gas formation such as trauma, gastrointestinal perforation, and gas-forming infection should also be considered. In rare cases, perforation of the gastrointestinal tract produces subcutaneous emphysema, especially that involving the lower extremities. We report a case of extensive subcutaneous emphysema of the left lower extremity following perforation of the rectum, initially diagnosed as GFM.

Case Report

A 73-year-old male patient had a history of rectal cancer with bladder adhesion and underwent post low anterior resection 10 months before visiting our ED. He received adjuvant combined chemotherapy and radiotherapy (CCRT) after the operation due to an initial lack of tissue proof for malignancy with a colonoscopic biopsy. He came to our ED complaining of sudden left lower leg pain and progressive swelling for two days. On physical examination, the whole left thigh, knee, leg, and ankle showed mild swelling without crepitus, bullae, or blister formation. Laboratory data showed leukocytosis (11,930/μL) with left shifting, elevated C-reactive protein (269.70 mg/L), and acute kidney injury (creatinine = 1.8 mg/dL). Unexpectedly, crepitus developed diffusely over the left

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buttock, thigh, leg, and ankle during the few hours the patient stayed in the ED's holding area. Computed tomography (CT) of the extremities was then performed with the suspension of GFM, revealing an infection process with diffuse gas formation in subcutaneous areas of the left buttock, thigh, and leg down to the foot (Fig. 1). Moreover, a few free air bubbles had gathered in the pelvic floor, which indicated that the gas had originated in the pelvic cavity. Further abdominal CT revealed gas-forming myofasciitis in the pelvic floor, with a suspected rectal perforation with abscess formation in the presacral area (Fig. 2). Proctoscopy accordingly revealed mucosal leakage in the rectal anastomosis site. Under the impression of rectum perforation with extensive GFM of the left lower limbs, the patient received an emergency fasciotomy and loop colostomy. Neither pus collection nor muscle necrosis was found during the operation, and only odor gas was drained. After five days, during incision and drainage, the colorectal surgeon found an anastomosis separation on the posterior side of the rectum, with some stool and dirty discharge inside the pelvis. Fasciotomy and fasciectomy were performed a further three times owing to poor wound healing and wound-edge necrosis. The patient was discharged after 43 days of hospitalization, in a stable condition.

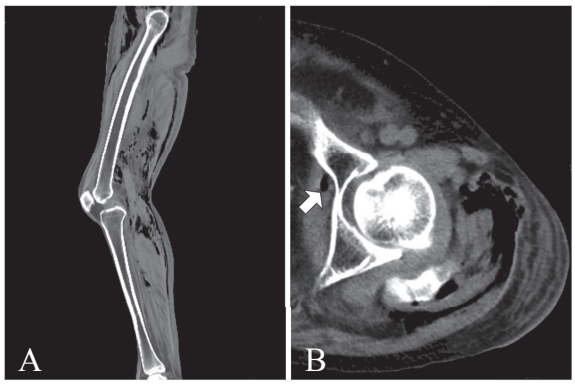

Fig. 1. Sagital view and axial view of extremities computed tomography (CT) without contrast. (A) It displayed extensive subcutaneous emphysema of left lower extremity mimic gas-forming myonecrosis (GFM). (B) It showed air existed in the pelvic cavity (arrow) provided a clue that the gas of the left lower extremities may originate from pelvic cavity.

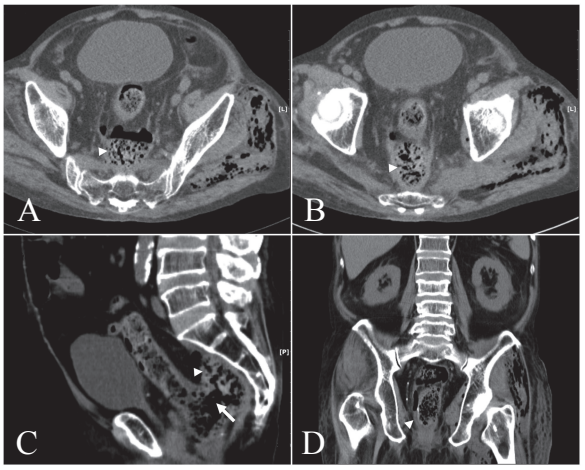

Fig. 2. (A–D) axial view, corona view and sagittal view of abdominal computed tomography (CT) without contrast. The figure showed air-containing perirectal abscess (arrow head) which communicate with pararectal space. (C) It displayed the posterior rectum wall was separated as well (arrow). The air dissected into pelvic floor with extension to the left lower extremity.

Discussion

Intra-abdominal disease is a rare cause of subcutaneous emphysema in the extremities. In the reported case, rectal perforation from an anastomosis site related to colon cancer surgery resulted in air dissecting to the subcutaneous and muscle layers of the left thigh and leg. The rapid and extensive development of crepitus in the subcutaneous area also indicated that the gas originated from the rectal perforation site rather than from GFM. The results of the initial fasciotomy confirmed this. Moreover, subcutaneous emphysema of the lower limb resulting from gastrointestinal perforation, which is a rare condition that could also mimic GFM or gas gangrene, required a differential diagnosis, which is difficult to perform in the ED.[1] The major mechanism of subcutaneous emphysema in this case was not bacterial but rather the pressure gradient between the lumen of the gut and the surrounding tissue.[2,3] The usual route of extravasation is directly through a pathological defect in the parietal peritoneum or fascia contiguous with this defect and into the intramuscular plane and subcutaneous spaces where infection may subsequently occur.[4,5] In the literature, we found one similar case in which rectal perforation had been diagnosed until some fecal material accumulated over the fasciotomy wound.[6] In our case,

quick diagnosis of rectal perforation was possible due to the few air bubbles detected on the extremity CT and the past history of colon cancer.[4] Some articles have suggested that performing a barium enema could confirm diagnosis of a bowel perforation.[7] In general, patients with colon cancer should receive CCRT before the operation. However, this patient underwent two biopsies without tissue proof for malignancy. Due to a strong suspicion of malignancy, he received low anterior resection directly before CCRT. This may have caused anastomosis breakdown and colon perforation in this patient.[8]

Extensive antibiotic coverage for mixed flora and anaerobic bacteria, especially gas-forming organisms, should be considered in such a case. Generally, patients with necrotizing soft tissue infections are treated with surgical intervention and antibiotics. For patients with subcutaneous emphysema of intra-abdominal origin in the extremities, an exploratory operation for lesions combined with a fasciotomy in the involved extremities is reasonable.[1]

Conclusions

When a patient presents with subcutaneous gas formation in the extremities, infection from an underlying abdominal cause should be considered in addition to local bacterial infection. Other cases have reported similar conditions originating from renal abscess, small bowel perforation, perirectal abscess, rectal perforation, and emphysematous cholecystitis (Table 1).[9] Examining all possible sources of infection is crucial.

Conflicts of Interest Statement

All the authors declared that there is also no conflict of interest regarding the publication of this article.

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All authors have no commercial association, such as consultancies, stock ownership or other equity interests or patent-licensing arrangements.

Ethics and Consent

This article is a case report which does not included human experiment and any privacy information of patient. Based on the Institute Reviewed Board policy and regulars, the Institute Reviewed Board did not need to oversee manuscripts of a case report. The identity of our patient has been removed and deleted carefully in this article to ensure the patient's privacy and rights.

Availability of Data and Materials

All data generated during this study are included in this published article.

Blinding

The response letter is properly blinded of authors' identity.

References

1. Hafez MA. Subcutaneous emphysema of the leg: could be a fatal condition. *Open J Clin Diagn* 2012;2:56-58. doi:10.4236/ojcd.2012.23012
2. Oetting HK, Kramer N, Branch W. Subcutaneous emphysema of gastrointestinal origin. *Am J Med* 1955;19:872-886. doi:10.1016/0002-9343(55)90155-8
3. Quigley J, Brody DA. A physiologic and clinical consideration of the pressures developed in the digestive tract. *Am J Med* 1952;13:73-81. doi:10.1016/0002-9343(52)90082-X
4. Jager GJ, Rijssen HV, Lamers JJ. Subcutaneous emphysema of the lower extremity of abdominal origin. *Gastrointest Radiol* 1990;15:253-258. doi:10.1007/BF01888788
5. Tan CH, Vikram R, Boonsirikamchai P, Faria SC, Charnsangavej C, Bhosale PR. Pathways of extrapelvic spread of pelvic disease: imaging findings. *Radiographics* 2011;31:117-133. doi:10.1148/rg.311105050
6. Ito T, Shiraki K, Sekoguchi K, et al. Case report: metastatic gas gangrene of the leg due to acute emphysematous cholecystitis. *Dig Dis Sci* 2001;46:2480-2483. doi:10.1023/A:1012336222483
7. Kountouras J, Zavos C. Recent advances in the management of radiation colitis. *World J Gastroenterol* 2008;14:7289-7301. doi:10.3748/wjg.14.7289
8. Lee KB, Moon ES, Jung ST, Seo HY. Subcutaneous emphysema mimicking gas gangrene following perforation of the rectum: a case report. *J Korean Med Sci* 2004;19:756-758. doi:10.3346/jkms.2004.19.5.756
9. Pickels RF, Karmody AM, Tsapogas MJ, Griffin P. Subcutaneous emphysema of the lower extremity of gas-

Table 1. The following case series including 10 cases since 1970's with subcutaneous emphysema of lower extremities resulted from abdominal origin in the literature reviews

Patient	Reference	Year	Age	Sex	Author	Clinical presentation	Diagnosis	Treatment	Outcome
1	9	1974	76	M	Pickels et al.	Back pain	Transverse colon diverticula communicated with psoas abscess	Antibiotics and fasciotomy	Death
2	10	1978	46	F	Fox et al.	Left hip pain	Rectosigmoid perforation	Antibiotics and fasciotomy	Resolution
3	4	1990	65	F	Jager et al.	Right hip pain	Perforated sigmoid with fistula	Antibiotics and colostomy	Death
4	4	1990	43	M	Jager et al.	Perineal pain	Perirectal abscess	Antibiotics and amputation	Death
5	4	1990	62	M	Jager et al.	Right upper leg pain	Nontraumatic metastatic gas gangrene due to emphysematous cholecystitis	ERCP and antibiotics	Death
6	6	2001	72	M	Ito et al.	Abdominal pain and followed with right leg pain	Acalculous emphysematous cholecystitis, subcutaneous and intramuscular emphysema of right upper leg extending to the thigh	PTGBD and antibiotics	Death
7	8	2004	38	M	Lee et al.	Right thigh pain	Rectal ulcer perforation	Low anterior resection and antibiotics	Resolution
8	11	2006	70	M	McMullin et al.	Fournier's gangrene	Perforated rectal carcinoma	Antibiotics and incisional drainage	Death
9	12	2010	64	M	Saldua et al.	Left hip pain and dysuria	Perforation of small intestine	Surgery, antibiotics, and repeat drainage	Resolution
10	1	2012	70	F	Hafez	Left leg pain	Left kidney staghorn calculus with abscess	Incisional drainage and antibiotics	Death

ERCP: endoscopic retrograde cholangiopancreatography; F: female; M: male; PTGBD: percutaneous transhepatic gallbladder drainage.

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trointestinal origin: report of a case. *Dis Colon Rectum* 1974;17:82-86. doi:10.1007/BF02587542

10. Fox TA, Gomez J, Bravo J. Subcutaneous emphysema of the lower extremity of gastrointestinal orgin. *Dis Colon Rectum* 1978;21:357-360. doi:10.1007/BF02586667
11. McMullin NR, Gering S, Levoyer T. Necrotizing Fournier's gangrene from a perforated rectal carcinoma. *Surg Rounds* 2006;29:275.
12. Saldua NS, Fellars TA, Covey DC. Case report: bowel perforation presenting as subcutaneous emphysema of the thigh. *Clin Orthop Relat Res* 2010;468:619-623. doi:10.1007/s11999-009-1015-3