Subcutaneous cervical emphysema and pneumomediastinum due to a diastatic rupture of the cecum

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SUMMARY: Subcutaneous cervical emphysema and pneumomediastinum due to a diastatic rupture of the cecum.

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Pneumomediastinum usually occurs after esophageal or chest trauma. Subcutaneous cervical emphysema as a presentation of non-traumatic colonic perforation following colorectal cancer or diverticulitis, is very rare.

We report a case of a patient with rectal cancer who developed a

diastatic cecum retroperitoneal perforation with a secondary pneumomediastinum and cervical emphysema. The patient was in treatment with a neoadjuvant chemo-radiotherapy for a low rectal cancer.

Treatment consisted in an emergency right hemi-colectomy with ileostomy and performance of distal colonic fistula.

The Authors discuss the occurrence of pneumomediastinum and cervical emphysema complicating rectal cancer, pointing out ethiopathogenesis, clinical presentation, diagnosis and treatment. The importance of performing a diverting colostomy when neoadjuvant chemotherapy is scheduled in patients with stenotic rectal cancer, although not clinically occluded

KEY WORDS: Pneumomediastinum - Cervical emphysema - Rectal cancer.

Introduction

Pneumomediastinum and subcutaneous emphysema due to colonic perforation are uncommon clinical entities. They have been sporadically described as complications of diverticulitis (1, 2), toxic megacolon in ulcerative colitis (3) and colonoscopy (4, 5) with endoscopic polypectomy (6).

Only few reports have been published in the Literature in patients with colo-rectal cancer. In the isolated cases reported in the Literature subcutaneous emphysema has been described in advanced colo-rectal cancer complicated by occlusion and retroperitoneal perforation of left colon.

We report a unique case of pneumomediastinum with

cervical subcutaneous emphysema, which occurred in a patient with unresectable advanced rectal cancer, who developed during neo-adjuvant chemio-radiotherapy a complete neoplastic obstruction with a diastasis retroperitoneal perforation of the caecum. Implications in the therapeutic oncologic strategy related to this rare observation are discussed. The usefulness of diverting colostomy in advanced rectal cancer scheduled for neoadjuvant oncologic therapy is debated according to this case report and review of the Literature.

Case report

A 58 years-old male patient was initially referred to our Department for a rectal cancer located nearly 6 cm from the anus at the colonoscopy. Total body CT-scan revealed metastasis to the omentum, the abdominal and thoracic lymph nodes, the liver and the lungs. MRI of the pelvis, performed in order to study the local diffusion of the disease, showed the locally advanced stage of the rectal cancer, which partially involved and obstructed the intestinal lumen. The cancer was considered unresectable, and the patient was referred to the oncologist. First line neo-adjuvant chemotherapy was started.

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Corresponding author: Eva Intagliata, e-mail: evaintagliata@vodafone.it © Copyright 2015, CIC Edizioni Internazionali, Roma Two months later, the patient presented poor clinical condition, with abdominal pain, lack of appetite and intestinal occlusion. At the physical examination, the abdomen was untreatable, tense and painful. At the abdominal X-ray, marked meteoric swelling with coarse airfluid levels were found. Soon after, the patient developed alteration of the voice, neck turgor and subcutaneous emphysema. Therefore, he was urgently referred to our Department of Surgery, and a thoracic and abdominal CT-scan was performed. The radiologic exam confirmed the huge intestinal swelling with signs of intestinal occlusion (Figure 1). CT-scan of the thorax revealed signs of pneumomediastinum without pleural effusion, and subcutaneous cervical emphysema (Figure 2). The patient was then operated in an emergency setting.

At laparotomy, a peritoneal fluid collection was found with multiple peritoneal metastasis and retroperitoneal diastatic perforation of the cecum. A right hemi-colectomy and an ileostomy with a distal colonic fistula without intraperitoneal anastomosis were accomplished.

The post-operative period was uneventful. Eight days later the patient was discharged from the hospital and referred to the oncologist in order to control his neoplastic disease.

Discussion

Pneumomediastinum and subcutaneous emphysema of the neck are uncommon clinical entities that occur when air leaks into the mediastinum from the lungs or any of the luminal organs of the chest.

Rarely, they occur after perforation of the luminal abdominal organs. Pneumomediastinum and subcutaneous emphysema have been described as a complication of colonic perforation mainly in patients with diverticulitis or toxic megacolon or in patients submitted to endoscopic procedures (1, 3-15). Only few occasional reports of pneumomediatinum have been published in patients with colo-rectal cancer (16-18).

The occurrence of pneumomediastinum and subcutaneous cervical emphysema in colon perforations may be explained considering the continuum of fascial planes connecting cervical soft tissues with the mediastinum and retroperitoneum (10, 21). When the perforation occurs in the extraperitoneal surface of the colon, the lumen contents and gas may proceed along the retroperitoneum, and may overtake the diaphragm, diffusing into the mediastinum. This diffusion is made possible since the spaces between the retroperitoneum, mediastinum and the cervical subcutaneous tissue are anatomically continuous. According to Maunder et al., the continuity is assured through the routes of subcutaneous tissue, prevertebral tissue, visceral space, previsceral space (21). Therefore, the escaped gas from the colon may dif-



Fig. 1 - Huge intestinal swelling with signs of intestinal occlusion.

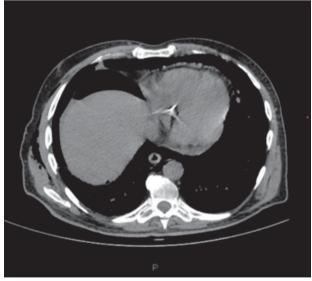


Fig. 2 - Pneumomediastinum and subcutaneous cervical emphysema.

fuse superiorly through the above-described anatomical routes into the mediastinum and then into the fascial areas, therefore causing pneumomediastinum and cervical subcutaneous emphysema.

In our case, the perforation occurred as a result of an obstructed unresectable rectal cancer, during the neoadjuvant chemotherapy. The chronic untreated occlusion caused the entire colon dilation. With a perfect continent ileo-cecal sphincter, the tight cancer stenosis caused a diastatic rupture of cecum. The anatomical site of perforation determined the direction of the air diffusion. Usually, the intestinal perforation happens into the abdominal cavity. In our case, perforation involved the posterior aspect of the cecum, explaining the upper diffusion of intestinal gases into the mediastinum and subcutaneous cervical spaces.

In pneumomediastinum, small bilateral pneumothoraxes may also appear as a consequence of a rupture of the mediastinal pleura, secondary to the increased mediastinal pressure.

Symptoms of pneumomediastinum and cervical emphysema may include severe chest and neck pain, dyspnea and dysphonia, while physical signs consist of subcutaneous crepitation on the neck and Hamman's sign (a crackling sound on auscultation congruent with the cardiac cycle). Fever and leukocytosis are other objective findings of perforation. Radiological diagnosis is based on chest and abdominal radiography and CT scan (22). Abdominal CT scan plays an important role in identifying an occult perforation and whole body CT allows to evaluate the extension of escaped gas to different body districts. Thus, radiological imaging plays also an important role in allowing an early diagnosis, a proper therapeutic planning and follow-up with a better outcome.

The risk of mediastinitis and septicemia in these patients is elevated, and an aggressive therapeutic approach is therefore required. Treatment is directed to solve the perforation through laparotomy or laparoscopy with removal of the site of perforation (22-25). If mediastinitis has occurred, treatment of this dangerous complication is required with antibiotic therapy or surgical drainage.

In our case, a laparotomy was performed and removal of the right colon with an ileostomy and a colonic distal fistula was achieved. It may be questioned if during the emergency surgery performed to overcome the perforation, a surgical treatment of the rectal disease should have been performed. In our opinion, it can be done if the tumor is not diffused. In our case, the patient had a metastatic disease with involvement of the peritoneal cavity, the liver and the lung. Therefore, a surgical resection of the tumor, although only palliative, was not justified.

Besides the unusual presentation of perforation, the reported case is very interesting since it occurred in a patient with rectal circumferential stenotic cancer during adjuvant chemotherapy. According to oncologic guidelines, a low rectal cancer should be treated surgically after neoadjuvant chemotherapy, unless there is a complication such as an intestinal occlusion. Although in our patient at the time when neoadjuvant chemotherapy was started a clinical occlusion was not observed, we question if an initial diverting colostomy or an endoscopic rectal stent, anyway, should or should not be proposed.

References

- Besic N, Zgajnar J, Kocijancic I. Pneumomediastinum, pneumopericardium and pneumoperitoneum caused by peridiverticulitis of the colon: report of a case. Dis Colon Rectum. 2004;47(5):766-8.
- Soliani G, Dominici M, Bergossi L, Basaglia E, Pauli S, Carcoforo P. Acute colon diverticulitis in multiple myeloma patient: an unusual presentation of a colonic perforation. Case report. Ann Ital Chir. 2002;73(6):643-6.
- Mogan GR, Sachar DB, Bauer J, Salky B, Janowitz HD. Toxic megacolon in ulcerative colitis complicated by pneumomediastinum: report of two cases. Gastroenterology. 1980;79(3):559-62.
- Goerg KJ, Duber C. Retroperitoneal, mediastinal and subcutaneous emphysema with pneumothorax after colonoscopy. Dtsch Med Wochenschr. 1996;121:693-6.
- Dehal A, Tessier D. Intraperitoneal and Extraperitoneal Colonic Perforation Following Diagnostic Colonoscopy JSLS. 2014;18(1):136-141.
- 6. Ho HC, Burchell S, Morris P, Yu M. Colon perforation, bilateral pneumothoraces, pneumopericardium, pneumomediastinum and subcutaneous emphysema complicating endoscopic polypectomy: anatomic and management considerations. Am

- Surg. 1996;62(9):770-4.
- Annahàzi A, Polyàk I, Nagy F, Wittmann T, Molnár T. "Ulcerative crepitus" A case with subcutaneous emphysema and pneumomediastinum without colonic perforation or toxic megacolon in ulcerative colitis successfully treated conservatively. Journal of Crohn's and Colitis. 2012;6(6):717-719.
- 8. Fosi S, Giuricin V. Subcutaneous Emphysema, Pneumomediastinum, Pneumoretroperitoneum and Pneumoscrotum: Unusual Complications of Acute Perforated Diverticulitis. Case Reports in Radiology. Volume 2014 Article ID 431563, 5 pages.
- Wiles T, Mullett R, Chadwick M. Diverticular perforation: an unusual cause of subcutaneous emphysema. Emerg Med J. 2014;31:259-260.
- Yasar N, Kebapci M, Ihtiyar E. Pneumomediastinum and subcutaneous emphysema caused by sigmoid diverticulum perforation secondary to blunt abdominal trauma: report of a case. Turkish Journal of Trauma and Emergency Surgery. 2011:93-95.
- 11. Koichiro S, Itoh S, Shigiyama F, Kitagawa T, Maetani I. Pneumoretroperitoneum, pneumomediastinum and subcutaneous emphysema after colorectal endoscopic submucosal dissection with air insufflation. J Gastroenterol. 2011;1(3):136-138.
- 12. Hur T, Chen Y, Shu GH, Chang JM, Cheng KC. Spontaneous

- cervical subcutaneous and mediastinal emphysema secondary to occult sigmoid diverticulitis. Eur Respir J. 1995;8:2188-2190.
- 13. Vecchio R, Marchese S, Leanza V, Leanza A, Intagliata E. Totally laparoscopic repair of an ileal and uterine iatrogenic perforation secondary to endometrial curettage. Int Surg. 2015 Feb;100(2):244-8.
- Vecchio R, MacFadyen BV. Laparoscopic common bile duct exploration. Langenbecks Arch Surg. 2002 Apr;387(1):45-54. Epub 2002 Apr 10. Review.
- 15. Vecchio R, Marchese S, Famoso S, La Corte F, Marletta S, Leanza G, Zanghì G, Leanza V, Intagliata E. Colorectal cancer in aged patients. Toward the routine treatment through laparoscopic surgical approach. G Chir. 2015 Jan-Feb;36(1):9-14.
- 16. Chu S, Glare P. Subcutaneous emphysema in advanced cancer. Journal of Pain and Symptom Management. 2000;19:73-79.
- 17. Chow E, Wong S, Goldberg RE, Stern HS. Non-traumatic subcutaneous emphysema in association with rectal carcinoma. Can Assoc Radiol J. 1996;47:94-97.
- Morita T, Matsuda T, Tei Y, Takada T. Nontraumatic Subcutaneous Emphysema from Rectal Cancer Perforation Completely Resolved After Intensive Pain Control. Letters j.jpainsmman.2006, 3-4.
- Schmidt G, Bronkhorst M, Hartgrink HH, Bouwman LH. Subcutaneous cervical emphysema and pneumomediastinum due to

- a lower gastrointestinal tract perforation. World J Gastroenterol. 2008;14(24):3922-3923.
- 20. Murariu D, Tatsuno B, Tom MK, You JS, Maldini G. Subcutaneous Emphysema, Pneumopericardium, Pneumomediastinum and Pneumoretroperitoneum Secondary to Sigmoid Perforation: a case report. Hwaii J Med Public Health. 2012;71(3):74-77.
- Maunder RJ, Pierson DJ, Hudson LD. Subcutaneous and mediastinal emphysema. Pathophysiology, diagnosis and management. Arch Intern Med. 1984;144(7):1447-53.
- Pretre R, Robert J, Mirescu D, Witzig JA, Rohner A. Pathophysiology, recognition and management of pneumoretroperitoneum. Br J Surg. 1993;80:1138-1140.
- Tewari S, Getrajdman G, Petre EN, Sofocleous CT, Siegelbaum RH, Erinjeri JP, Weiser MR, Thornton RH. Safety and Efficacy of Percutaneous Cecostomy/Colostomy for Treatment of Large Bowel Obstruction in Adults with Cancer. JVIR. 2015:182-188.
- Vecchio R, Intagliata E, La Corte F, Marchese S, Cacciola RR, Cacciola E. Late results after splenectomy in adult idiopathic thrombocytopenic purpura. JSLS. 2015 Jan-Mar;19(1):e2013. 00272. doi: 10.4293/JSLS.2013.00272. PMID:25848175.
- Vecchio R, Cacciola E, Martino M, Cacciola RR, MacFadyen BV. Modifications of coagulation and fibrinolytic parameters in laparoscopic cholecystectomy. Surg Endosc. 2003 Mar;17(3):428-33