

A Case of Pneumorrhachis and Pneumoscrotum Following Colon Endoscopic Submucosal Dissection

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Endoscopic submucosal dissection has been a useful treatment of selected colorectal neoplasia cases. The incidence of perforation related to colorectal endoscopic submucosal dissection is 5-20%. However, while there have been numerous reports regarding retroperitoneal, mediastinal, pleural and subcutaneous emphysema after therapeutic colonoscopy, pneumoscrotum is a relatively rare manifestation of perforation associated with colorectal endoscopic submucosal dissection. In particular, pneumorrhachis, or air within the spinal cord, following therapeutic colonoscopy, is extremely rare. Herein, we report a conservatively treated perforation case as having pneumorrhachis, pneumoscrotum, and pneumoperitoneum after colorectal endoscopic submucosal dissection. (**Intest Res 2013;11:208-212**)

Key Words: Pneumorrhachis; Pneumoscrotum; Colon perforation; Endoscopic submucosal dissection

INTRODUCTION

Endoscopic submucosal dissection (ESD) is a useful therapeutic technique for the selected cases of colorectal tumor enabling en bloc resection regardless of the tumor size. While colorectal ESD is effective and less invasive compared to surgery, it has not been widely performed because of technical difficulties and perforation rate of approximately 5-20%.¹⁻³ However, many medical centers in Korea have recently started performing colorectal ESD through technical refinement and development of new devices and equipment. Several specialized referral centers in Japan and other Asian countries have also recently performed colorectal ESD as one standardized technique.⁴ While there have been numerous reports of retroperitoneal, mediastinal, pleural and subcutaneous emphysema after therapeutic colonoscopy,⁵⁻⁷ pneumoscrotum has been a relatively rare manifestation of perforation associated with therapeutic colonoscopy.⁸ In particular, pneumorrhachis, following colonoscopy, is extremely

rare. In this paper, we report a case of pneumorrhachis and systemic subcutaneous emphysema, including pneumoscrotum, following perforation during colorectal ESD.

CASE REPORT

A 61-year-old man was referred to the hospital for treatment of a laterally spreading tumor (LST) 4 cm in diameter, which was located in the sigmoid colon 24 cm from the anal verge (Fig. 1). It was decided to treat the tumor using ESD without preprocedural histologic diagnosis because the tumor was granular-type suspicious of adenoma or adenocarcinoma confined to the mucosa.

ESD was performed using gastroscope (GIF-Q260; Olympus, Tokyo, Japan) with room air insufflation. Following elevation of the submucosal layer by injection of a sodium hyaluronate acid and saline mixed solution (Fig. 2A), circumferential incision and submucosal dissection were performed, using a flex knife and IT-II knife (Olympus). The tissue was dissected along the submucosal layer after incising approximately one-third of the lesion. Thirty minutes after initiating the procedure, a small perforation occurred during dissection using a flex knife (Fig. 2B). During the 10 minutes following perforation, dissection of the surrounding area was done before endoclippping because the clips could

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have disturbed submucosal dissection. The perforation was then closed successfully using endoclipping (Fig. 2C). ESD could be continued because the patient did not complain of any significant symptoms, which might be related with perforation during the procedure, and finally, the tumor was removed in en bloc fashion. The specimen size was 48×38 mm,



Fig. 1. Endoscopic view of laterally spreading tumor, located in sigmoid colon 24 cm from the anal verge.

and the lesion was 42×30 mm (Fig. 2D). The duration of the procedure after endoclipping was 80 minutes.

Immediately following the procedure, the patient had no abdominal symptoms, but complained of chest discomfort with swelling over his neck and left anterior chest. Physical examination revealed subcutaneous emphysema of the neck, thorax, abdomen, hip, scrotum and left lower extremity. Radiograph showed left pneumothorax, pneumomediastinum, pneumoperitoneum, pneumoretroperitoneum and subcutaneous emphysema (Fig. 3). CT of the abdomen and pelvis demonstrated pneumoscrotum, and air in the perineural space at the sacral cauda equine (Fig. 4). However, he had no signs or symptoms suggestive of pneumorrhachis or pneumoscrotum.

Tube drainage of the left pleural cavity was performed because the pneumothorax did not respond to the empirical O₂ therapy, and it was aggravated the following day. The patient had no other symptoms, except left chest wall pain due to tube drainage. Therefore, the patient was managed conservatively with broad-spectrum antibiotics. The removed specimen was histologically diagnosed as a tubular adenoma with low-grade dysplasia. The emphysema gradually subsided and the patient was discharged on the 7th day after ESD. One month later, a follow-up CT revealed no abnormally leaked air.

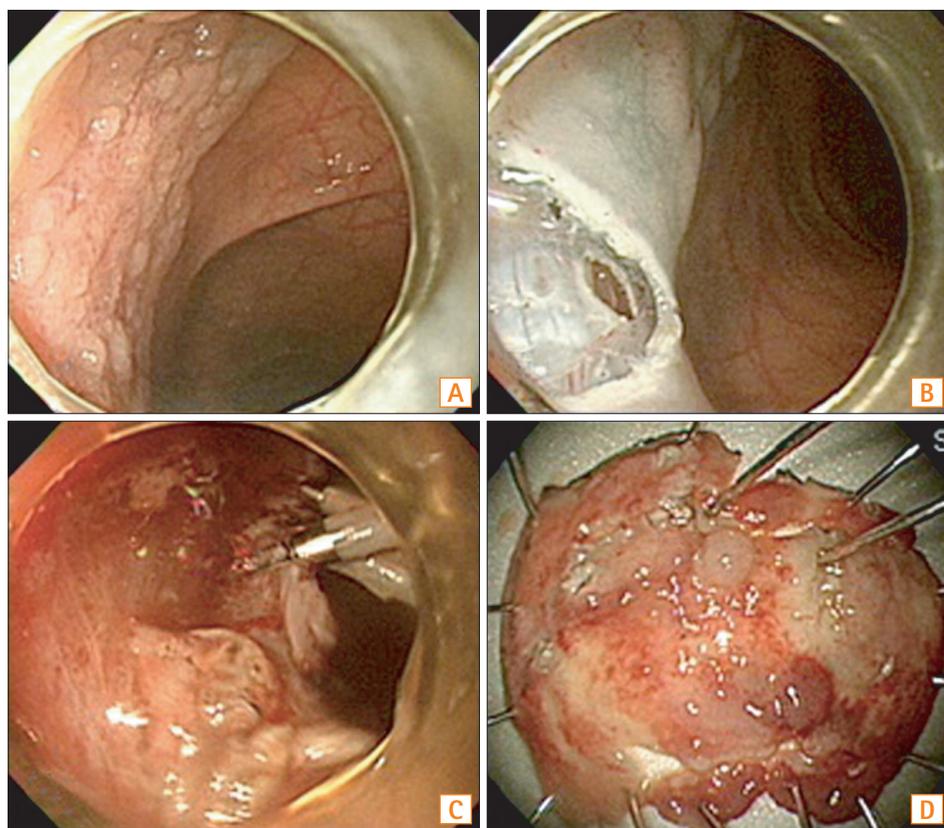


Fig. 2. Endoscopic resection of laterally spreading tumor of colon. (A) Lifting up the submucosal layer by injection of sodium hyaluronate acid and saline mixed solution. (B) Endoscopic view of submucosal fibrosis and small perforation. (C) Closure with endoclips due to perforation during endoscopic submucosal dissection. (D) Grossly resected specimen.

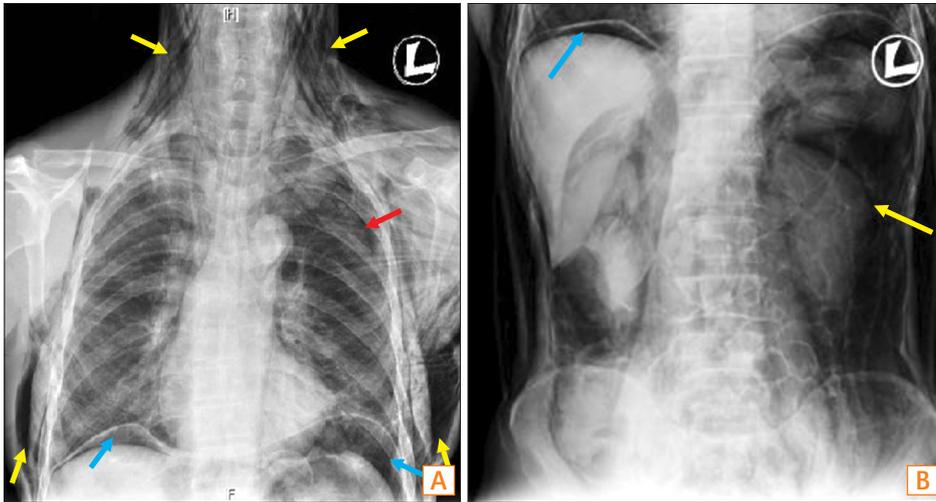


Fig. 3. (A) Chest radiograph showing subcutaneous emphysema (yellow arrows), left pneumothorax (red arrow) and free air in intraperitoneum (blue arrows). (B) Abdomen radiograph showing pneumoretroperitoneum (yellow arrow) and pneumoperitoneum (blue arrow).

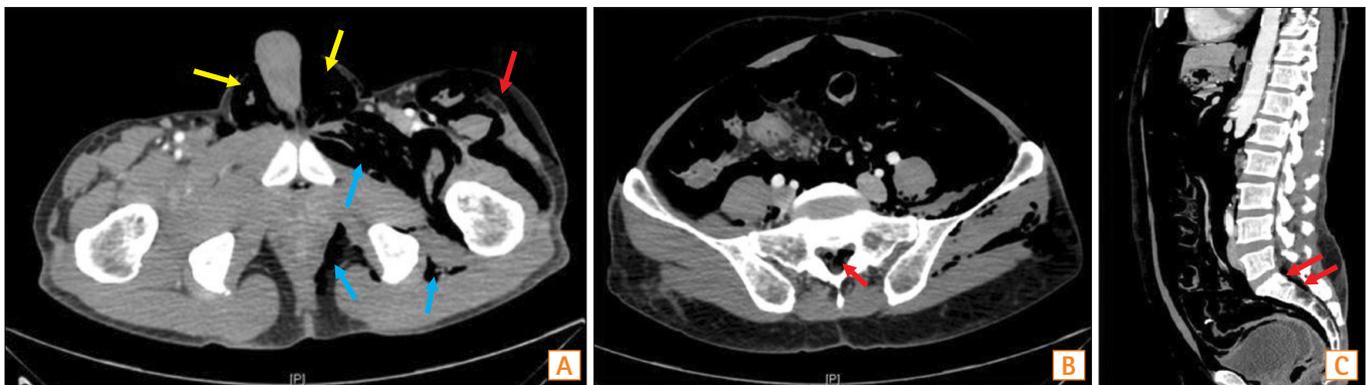


Fig. 4. (A) CT of abdomen and pelvis showing air in scrotum (yellow arrows), pelvis subcutaneous (red arrow) and intramuscular emphysema (blue arrows). (B, C) CT scan showing air within spinal canal (red arrows) at level of sacral cauda equina.

DISCUSSION

Although there have been numerous reports of retroperitoneal, mediastinal, pleural and subcutaneous emphysema after therapeutic colonoscopy,⁵⁻⁷ pneumosrotum is still regarded as a rare manifestation of perforation-associated colonoscopy. Its occurrence follows retroperitoneal perforation and can also be combined with pneumoperitoneum (Table 1).⁸⁻¹² Anatomically, the retroperitoneum communicates with the mediastinum and spermatic cord inferiorly and is continuous with the preperitoneal interstitium, while the mediastinum communicates with the bronchovascular interstitium and subcutaneous tissue. Therefore, air arising from the lesion in any of these areas can migrate into another region along the fascial planes.¹³

Pneumorrhachis is defined as the presence of air in the epidural or subarachnoid space. Retroperitoneal air may migrate along the fascial planes from the posterior mediastinum, through the neural foramina, and along the vascular and nerve root sheaths, and into the epidural space. Since intra-

spinal air is usually asymptomatic, diagnostic work-up should include spinal CT, the imaging tool of choice.¹⁴ Nontraumatic pneumorrhachis, in conditions that produce high intrathoracic pressure and barotraumas, have been described following spontaneous or non-trauma-related pneumomediastinum or pneumothorax.^{15,16} Nontraumatic pneumorrhachis is rare and even more so in cases associated with therapeutic colonoscopy.

Cases of pneumoretroperitoneum, pneumomediastinum, subcutaneous emphysema and pneumothorax, following colon perforation, have been treated conservatively or surgically.⁵⁻⁷ Up to now, criteria for non-surgical treatment of perforation during colorectal ESD remain unclear. Reported criteria by Hotta et al.¹⁷ were absence of diffuse peritonitis and successful closure with hemoclips. Patients with pneumosrotum were managed conservatively, except one case treated with laparotomy (Table 1).⁸⁻¹² Pneumorrhachis does not tend to migrate and reabsorbs spontaneously. Therefore, patients with pneumorrhachis are usually managed conservatively.¹⁴

Table 1. Review of Pneumoserotum Associated with Colonoscopy

Author	Age/gender	Procedure	Symptom	Diagnosis	Combined problem	Treatment (HD days)
Graepler et al. ⁸	65/male	Sigmoid polypectomy	Scrotal swelling	Plain radiograph	Pneumoperitoneum Subcutaneous emphysema	Clipping laparotomy (-)
Fishman and Goldman ⁹	75/male	Splenic flexure polypectomy	Scrotal pain and fullness	Plain radiograph	Pneumoperitoneum Pneumomediastinum	Conservative therapy (7)
Humphreys et al. ¹⁰	71/male	Sigmoid polypectomy	Scrotal swelling	Plain radiograph	Pneumoperitoneum Pneumomediastinum Subcutaneous emphysema	Conservative therapy (5)
Goerg and Düber ¹¹	25/male	Colonoscopy with biopsy in patient of severe UC	Scrotal swelling	Plain radiograph	Pneumoretroperitoneum Subcutaneous emphysema Pneumomediastinum Pneumothorax	Conservative therapy (7)
Fu et al. ¹²	52/male	EMR with APC for descending colon LST	Inguinal pain Scrotal swelling Mild fever	Plain radiograph CT	Pneumoperitoneum Pneumopericardium	Conservative therapy (5)

HD, hospital day; EMR, endoscopic mucosal resection; APC, argon plasma coagulation; LST, laterally spreading tumor.

Carbon dioxide (CO₂) insufflation, instead of room air, is safe and effective for patient comfort.¹⁸ Maeda et al.¹⁹ suggested insufflation of CO₂, rather than air during esophageal ESD reduced postprocedural mediastinal emphysema. While our case received room air insufflation, he was discharged on the 7th day after ESD with no sequelae. CO₂ insufflation, rather than room air, may have reduced procedure-related abdominal discomfort, but most likely would not have substantially changed the number of hospital-stay days, considering patients without complication after colorectal ESD are typically discharged on the 5th post-procedural day in our institution. In addition, use of the IT-knife is not widely accepted in colorectal ESD because of its technical difficulty and risk of complication, such as perforation and bleeding.^{2,20} However, in our experience, the IT-II knife can be used effectively for ESD of large colorectal LST, rather than other electrosurgical knives limited to the experts. Furthermore, perforation occurred during dissection using the flex knife because of submucosal fibrosis in this case. Further studies are needed of the use of CO₂ insufflation and appropriate knives for colorectal ESD.

Our report represents a rare case of pneumorrhachis and pneumoserotum secondary to pneumoretroperitoneum, following colorectal perforation during ESD. Close observation of signs and symptoms along with radiography enabled early diagnosis, and conservative measures produced a successful outcome.

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