Int J Clin Exp Pathol 2014;7(9):6345-6349
www.ijcep.com /ISSN:1936-2625/IJCEP0001453

Case Report

Squamous cell carcinoma of the small bowel manifesting as a jejunal perforation: a case report

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Received July 18, 2014; Accepted August 23, 2014; Epub August 15, 2014; Published September 1, 2014

Abstract: Squamous cell carcinoma arising from the small intestine is rare and difficult to identify as a primary or metastatic feature. We report a case of small intestinal squamous cell carcinoma manifesting as subacute peritonitis due to perforation. An 80-year-old man was admitted to our hospital with intermittent postprandial abdominal pain. He was diagnosed with acute peritonitis due to gastrointestinal perforation. During explorative laparotomy, a perforation site was detected in the jejunum and segmental resection to correct the perforation was performed including the perforation site located at the 70 cm inside the jejunum from the Treitz ligament. The pathology results revealed squamous cell carcinoma in the resected segment of the jejunum with two perforation sites.

Keywords: Squamous cell carcinoma, small intestine, jejunum, perforation, peritonitis

Introduction

Malignant tumors of the small intestine (SI) are very rare compared to those of other gastrointestinal organs; fewer than 2% of all GI malignancies originate in the small bowel [1, 2]. However, metastatic cancers of the small bowel are more frequent than primary SI cancers [3, 4]. Considering the pathologic types of those malignant tumors of the SI, approximately 30% to 50% are adenocarcinomas, 25% to 30% are carcinoids, and 15% to 20% are lymphomas [2]. Squamous cell carcinomas of the SI, either primary or metastatic, are exceptionally rare with only a few case reports in the literature [5-13]. We report a case of a pure squamous cell carcinoma arising in the jejunum manifesting as a small bowel perforation in an 80-year-old male.

Case report

An 80-year-old male patient who had been diagnosed with diabetes, hypertension and end-stage renal disease presented complaining of intermittent postprandial abdominal cramps for five days. He did not complain of nausea, vomiting, constipation, or diarrhea. His blood pressure was 151/71 mm Hg with a heart rate of 88 beats/min, a body temperature of 38.0°C, and a SpO_2 of 93.1% in room air. On the initial chest X-ray, we found diffuse patchy infiltrations and increased interstitial markings in both lung fields that were located around the trachea and major bronchi, with no evidence of a mass-like lesion (**Figure 1**). No palpable lymphadenopathy was noted during the physical examination of the head and neck region. However, on the physical examination, there were marked abdominal tenderness and peritoneal irritation and there was diffuse gas and fluid-loading in small and large bowel loops, suggesting paralytic ileus, on the abdominal x-ray. His laboratory tests revealed white blood cell count 4,430 cells/m^3, hemoglobin of 6.2 g/dl, platelet level 137×10^5/L, albumin level 3.0 g/dl, blood urea nitrogen (BUN) level 58.3 mg/dl, creatinine (Cr) level of 5.90 mg/dl, and C-reactive protein (CRP) level of 2.01 mg/dl. His aspartate aminotransferase (AST) and alanine aminotransferase (ALT) levels were normal and prothrombin time international normalized ratio (PT-INR) was 1.22 (1.5-2.5). An abdominal computed tomography (CT) scan revealed free air and massive ascites in the Douglas fossa (**Figure 2**). Although the site of perforation was not identified, a clinical diagnosis of acute peritonitis with gastrointestinal perforation was made and an exploratory laparotomy was conducted.

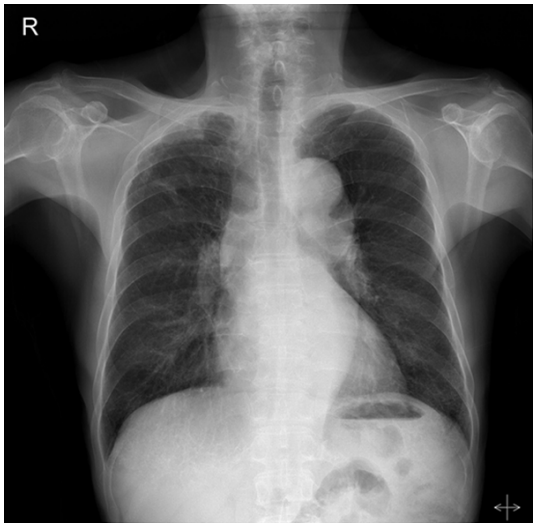

Figure 1. The initial chest X-ray showed diffuse patchy infiltrations and increased interstitial markings in both lung fields with no evidence of a mass-like lesion.

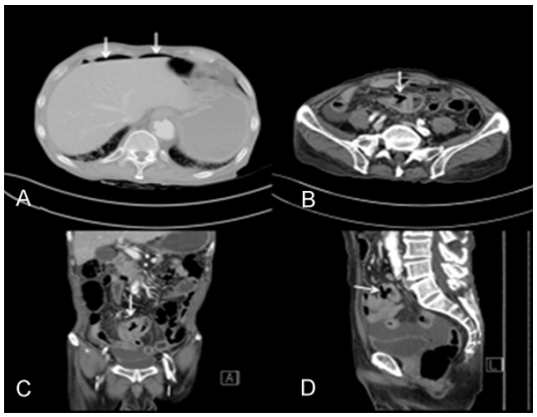

Figure 2. An abdominal CT scan revealed free air (pneumo-peritoneum; arrows in A), suggesting a hallow viscus (bowel) perforation. There were diffuse thickened distal jejunal loops with focal deformity (arrows in B, C and D) and a large amount of ascites with peritoneal wall enhancement in the Douglas pouch and paracolic gutter.

During surgical exploration, ascites was found. In addition, a perforation was located 70 cm inside the jejunum from the Treitz ligament (**Figure 2**). An intestinal segmental resection of approximately 13 cm including the perforation site was performed. Washing of the intraperitoneal cavity with 6,000 ml saline solution and an end-to-end intestinal anastomosis was performed. After the resection, his urine output did not improve and postoperative follow-up chest

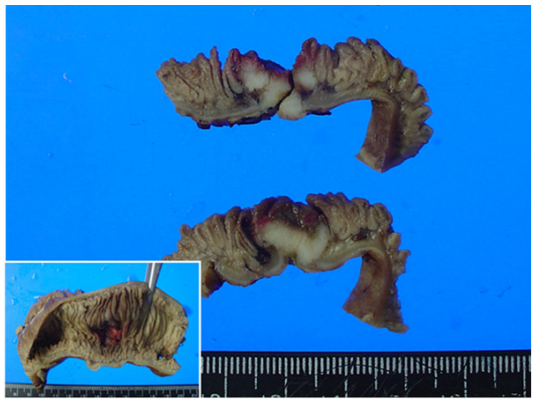

Figure 3. Grossly, in the resected jejunal segment, two perforations were noted: one on the center of the tumor 0.2 cm and 0.4 cm in diameter and the other located 4 cm from the tumor perforation site (*inlet*) and the tumor had infiltrated the full thickness of the wall and penetrated the serosa.

x-rays showed gradual pulmonary edematous change. On postoperative day (POD) 4, the AST/ALT increased from 29/13 U/L to 2059/556 U/L, BUN/Cr was elevated to 62.2/6.34 mg/dL, and the average urine output dropped to below 500 ml/day. He complained of aggravating dyspnea, and the chest X-ray showed increased haziness on both lung hilar regions, indicating an edematous change. An echocardiogram was performed and the ejection fraction was 27~31%. After consulting with nephrologists, continuous renal replacement therapy (CRRT) was administered on POD 23.

Despite supportive care in the intensive care unit and proper management, the patient's renal function, urine output, and pulmonary edema signs on the chest X-ray deteriorated. In addition, along with aggravation of dyspnea, sudden atrial flutter and hypoxia, and his blood pressure started to drop gradually. Consequently, the patient expired with multi-organ failure on POD 24.

Pathology

On urgent exploration laparotomy, an approximately 2.1×2.0-cm-sized ulceroinfiltrative solid tumor was found in the jejunum, located 70 cm from the Treitz ligament. At the center of the tumor, a perforation 0.2 cm in diameter was noted and another perforation, a 0.4-cm-diameter hole, was also found 4 cm from the tumor perforation site (**Figure 3**). Microscopically, the

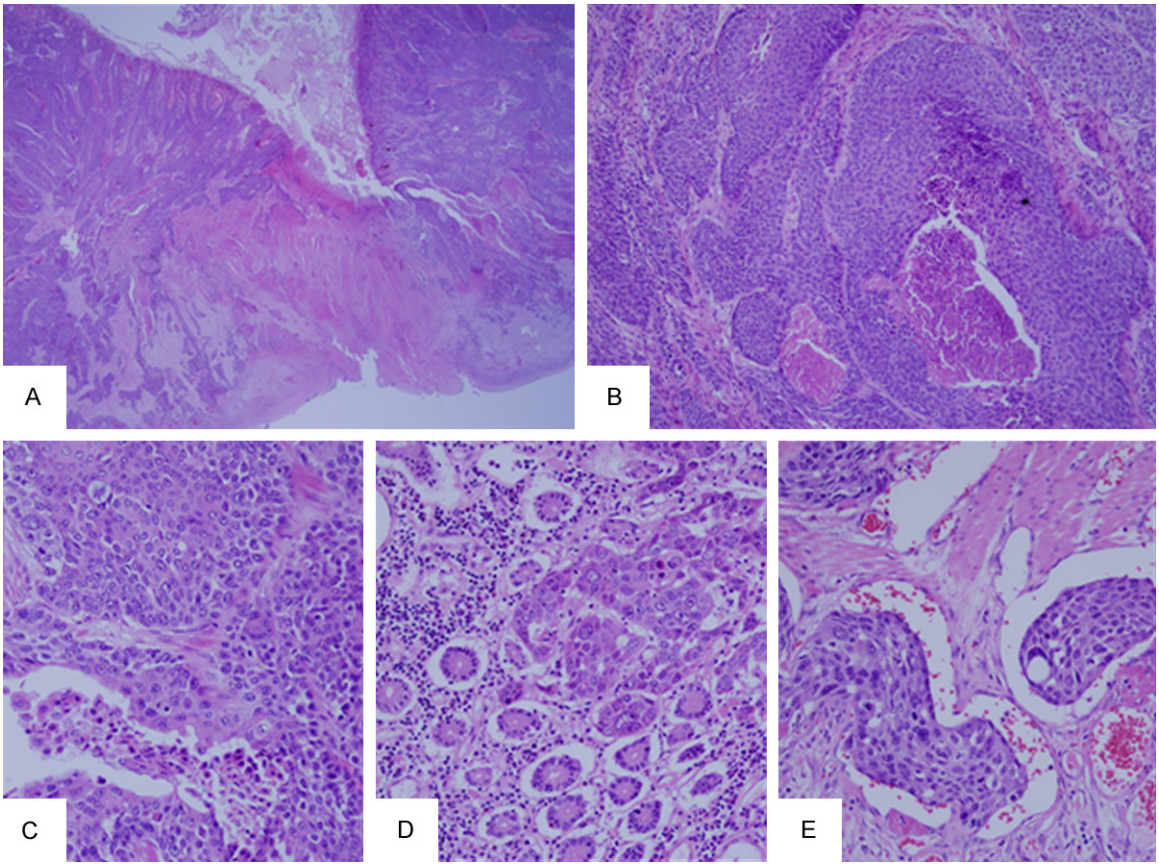

Figure 4. Microscopic findings of the tumor. A: Note that the tumor also showed a trans-mural tumor infiltration and serosal penetration, microscopically. B: Microscopically, the tumor consisted of well-demarcated nests and strands of large typical polygonal squamous cells and intercellular bridges. C: Most of the tumor cells showed prominent squamous differentiation, such as individual cell keratinization and intercellular bridges. D: Multiple foci of squamous metaplasia were seen in the normal glandular epithelium adjacent to the tumor. E: Numerous peritumoral lymphatic invasions were noted. (Hematoxylin-eosin stain, original magnifications, A, scan view; B, ×100; C-E, ×200).

tumor had infiltrated the full thickness of the wall and penetrated the serosa (**Figure 4A**). The tumor cells showed well-demarcated nests and strands architecture and composed of large typical polygonal squamous cells with prominent intercellular bridges (**Figure 4B**). Most of the tumor cells showed prominent squamous differentiation, such as individual cell keratinization and intercellular bridges (**Figure 4C**). Glandular differentiations were not found in any of the sections examined. Neither intracellular nor extracellular mucin was detected in the tumor with periodic acid-Schiff (PAS) reaction. Multiple foci of squamous metaplasia were seen in the normal glandular epithelium adjacent to the tumor (**Figure 4D**). Numerous lymphatic invasions were noted (**Figure 4E**). A diagnosis of infiltrating squamous cell carcinoma was made.

Discussion

Malignant tumors of the SI are very rare compared to those of other gastrointestinal organs [1, 2]. Among SI malignancies, squamous cell carcinomas (SCCs) are even more rare [11]. Only 3 cases (0.2%) of primary SCC were found in a recent analysis of 1.312 consecutive pathologic specimens of SI [11]. A review of the English literature revealed only three cases of squamous cell carcinoma of the small intestine (2 cases in the duodenum [7, 13], 1 case in the terminal ileum [12]). More commonly, squamous cell carcinomas may represent metastases [3, 4]. On autopsy, secondary tumors of the SI are 2.5 times more common than primary SI carcinomas [3]. The most frequent tumors metastasizing to the SI include carcinomas of lung, breast, kidneys and colon; malignant melanoma also commonly metastasizes to the SI

[4, 10, 14]. Garwood et al. reviewed 98 cases of SI perforation due to metastatic lung cancer and found that perforations occurred most often in the jejunum (53%) and that SI perforations were most often caused by adenocarcinoma (23.7%), squamous cell carcinoma (22.7%), large cell carcinoma (20.6%), and small cell carcinoma (19.6%) [15]. In another review of SI perforation, of the 19 cases, 4 cases were squamous cell carcinoma in which one primary site was the cervix, and 3 sites were from an unknown origin [6]. In the present case, the studies were limited due to the rapidly aggravating clinical course after resection of the SI and it was too risky to search for other possible primary sites by means of imaging tools on supposed SCC-originating organs. However, other evaluation techniques including chest X-ray, abdominal CT, and physical examinations were used and no suspicious primary squamous cell carcinoma originating sites other than the SI were found.

Pathologically, it is not easy to determine whether a SCC is primary or metastatic when it is found in the SI. Mumtaz et al. emphasized two useful differential points, in a case report on SCC arising in the ileum of a 65-year-old woman [9]. First, squamous metaplasia of the glandular epithelium favors it being the primary origin site if the majority of the cancer cells are deep in the wall of the SI. Second, if there is little involvement of the mucosa, the lesion is more likely to be metastatic [9]. However, Estrella et al. reviewed 100 metastatic carcinomas involving gastrointestinal (GI) mucosa and found that metastatic carcinomas involving the mucosal surface of the intestines have a tendency to exhibit gross and histologic features mimicking second primary tumors, especially when they originate from the GI tract [16]. And they concluded that *in situ* growth cannot be taken as prima facie evidence of a primary neoplasm.

In our case, the tumor showed prominent squamous differentiation, such as keratinization and intercellular bridges without glandular differentiation, such as glandular formation and intra- or extra- cellular mucin. Although the histopathologic diagnosis of infiltrating SCC was distinct, whether it was primary or metastatic was difficult to determine. First, our patient had no history of SCC, including of the lung, head or neck. In addition, on microscopic examination several foci of squamous metaplasia in the glandular epithelium adjacent to the tumor were noted. The evidence mentioned above indicated that the SCC of the SI in the present case may be of a primary SI carcinoma rather than metastatic.

In patients with either primary or metastatic small bowel tumors, acute abdominal symptoms should be regarded as a warning sign of perforation. Surgeons should be aware of the disease and decide early surgical intervention, which might increase the chances of survival in patients with perforation of small bowel tumors. Nevertheless, in the present case, the underlying diseases including diabetes, hypertension and renal insufficiency, may have been the leading cause of the fatal clinical course.

Disclosure of conflict of interest

None.

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