Case Report

Breast Signet-ring Cell Lobular Carcinoma Presenting with Duodenal Obstruction and Acute Pancreatitis

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We report here an extremely rare case of breast signet-ring cell carcinoma (SRCC) initially manifesting as duodenal metastasis and acute pancreatitis. A 62-year-old female presented with duodenal obstruction and swollen head of the pancreas, and the diagnosis of acute pancreatitis was initially made. Upper gastrointestinal endoscopy revealed duodenal stenosis with erosive mucosa, with signet-ring cells infiltrating the submucosal layer, suggesting duodenal metastasis of SRCC. Despite absence of a palpable mass in both breasts, computed tomography revealed diffuse enhancement of the left breast in addition to left axillary lymphadenopathy. Histological examination of mammary needle biopsy samples revealed SRCC with a non-invasive lobular carcinoma component. Primary breast SRCC with duodenal metastasis was therefore diagnosed. The patient underwent palliative surgery twice for intestinal obstruction due to peritoneal dissemination. She has remained alive without bowel obstruction for 18 months while being treated with cytotoxic chemotherapies. [*Asian J Surg* 2007;30(3):220–3]

Key Words: breast cancer, gastrointestinal tract, invasive lobular carcinoma, metastasis, peritoneal dissemination

Introduction

Signet-ring cell carcinoma (SRCC) can arise from various intra- and extra-abdominal organs. However, primary breast SRCC is rare, as is clinically diagnosed gastrointestinal (GI) metastasis from breast cancer. Most GI metastases from breast cancer occur as a part of systemic dissemination with multiple organ metastases, and such GI metastases are not rare in breast cancer autopsy cases.[1] However, there are few reports of symptoms of GI metastasis as an initial clinical sign of breast cancer.[2,3]

We report here an extremely rare case of non-palpable breast SRCC initially manifesting as duodenal obstruction and acute pancreatitis due to duodenal metastasis.

Case report

A 62-year-old female was admitted with postprandial nausea and vomiting of food residue over a 2-week period. Upper GI radiography, using water-soluble contrast medium, revealed obstruction of the descending portion of the duodenum. Abdominal computed tomography (CT) revealed a dilated pancreatic duct, swollen pancreatic head, and thick duodenal wall (Figure 1). Both plasma and uric amylase levels were high, suggestive of acute pancreatitis with duodenal obstruction. Upper GI endoscopy revealed stenosis of the duodenum with erosive mucosa. Biopsy samples obtained from the region of stenosis revealed signet-ring cells infiltrating the submucosal layer

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Figure 1. Abdominal computed tomography shows a swollen pancreas head (white arrow) and thick duodenal wall (arrowhead).

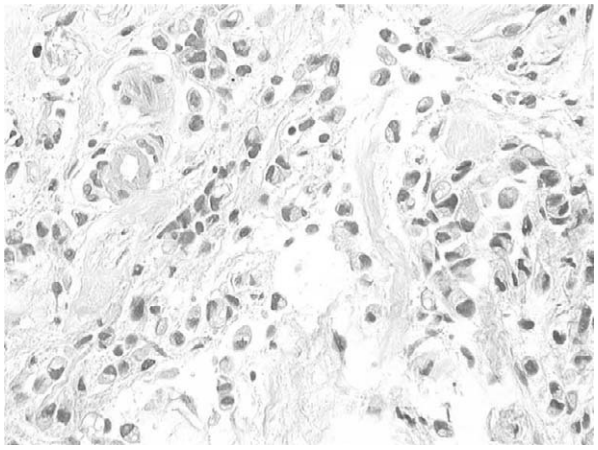

Figure 2. Histological examination of the duodenal biopsy specimen shows cancer cells infiltrating the submucosal layer with an intact mucosa; most of the cancer cells are signet-ring cells.

with an intact mucosa (Figure 2), suggesting duodenal metastasis of SRCC. Thereafter, to detect the primary lesion, neck and chest CT was performed and revealed diffuse enhancement of the left breast relative to the right, left axillary lymphadenopathy, and an osteolytic lesion in a thoracic vertebra, suggesting bone metastasis. No mass was palpated nor was any detected by ultrasound examination in the left breast. Mammography revealed architectural distortion in the outer part of the left breast. Mammary needle biopsy blindly aimed at the lesion revealed infiltration of SRCC with a noninvasive lobular carcinoma component (Figure 3). Immunohistochemically, ER/PR status, human epidermal growth factor receptor-2, and E-cadherin were negative in duodenal and breast biopsy specimens. Based on the histological and immunohistochemical similarities between the breast and duodenal lesions, primary SRCC of the left breast with bone and duodenal metastases was diagnosed.

At laparotomy, a 2-cm mass was found in the second portion of the duodenum. The pancreas was firm and swollen, indicating the presence of pancreatitis. Numerous small white nodules were noted in the peritoneum, indicating peritoneal carcinomatosis. Based on the presence of extensive dissemination, we performed gastrojejunostomy for palliation. Two weeks later, cytotoxic chemotherapy with paclitaxel was started. However, intestinal obstruction developed 2 months after the initiation of chemotherapy due to peritoneal dissemination, suggesting lack of favorable response to paclitaxel. Laparotomy was performed again, and partial resection of the ileum and transverse colostomy were performed for obstruction of the ileum and descending colon due to disease dissemination. Thereafter, anthracycline-based chemotherapy followed by docetaxel was initiated and is currently being continued. Two months after initiation of this chemotherapy, CT revealed that the swollen pancreatic head had normalized in size. The patient has remained alive without bowel obstruction for 18 months since first admission.

Discussion

Breast SRCC is a rare, aggressive variant that can originate from both invasive ductal and invasive lobular carcinoma (ILC).[4] In the present case, the presence of a noninvasive lobular carcinoma component indicated ILC as the origin of the SRCC, and enabled primary breast SRCC to be distinguished from metastasis of the breast from other organs. Clinicopathological differences between lobular and ductal carcinomas have been documented, including multicentricity and bilaterality in lobular carcinoma.[5] Among such differences, GI metastasis tends to occur with ILC.[6] In addition, SRCC exhibits a propensity to involve the GI tract.[7] Most GI metastases have been found at recurrence or at autopsy.[1] There have been few reports of GI manifestations preceding the diagnosis of breast cancer,[2] particularly for duodenal metastasis.[3] Acute pancreatitis induced by metastasis from lung or gastric cancer has been reported.[8] This is the first case, to our knowledge, of acute pancreatitis induced by breast cancer metastasis.

The mechanisms of induction of acute pancreatitis by metastasis include pancreatic ductal obstruction by metastatic tumour in the pancreas or peripancreatic lymph

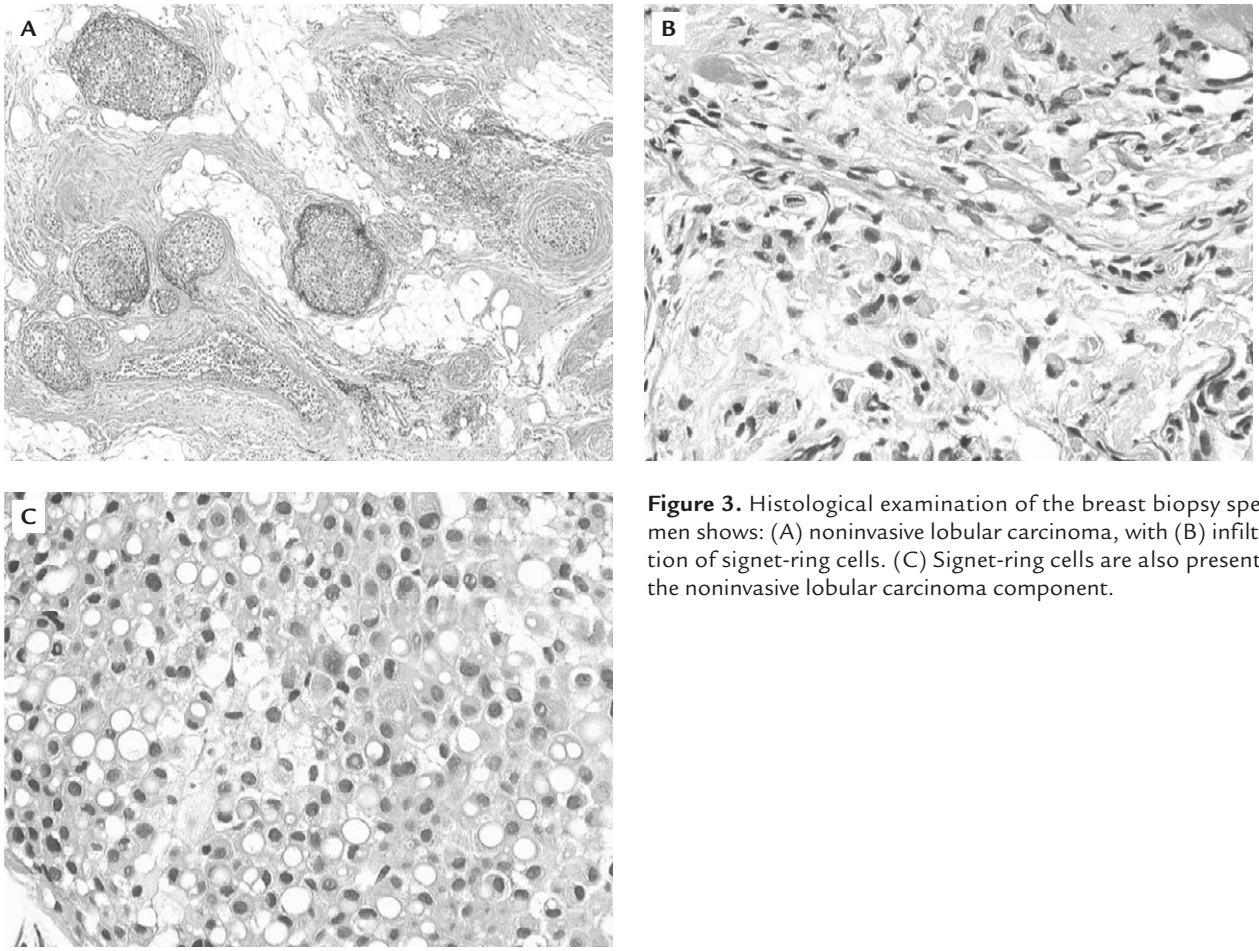

Figure 3. Histological examination of the breast biopsy specimen shows: (A) noninvasive lobular carcinoma, with (B) infiltration of signet-ring cells. (C) Signet-ring cells are also present in the noninvasive lobular carcinoma component.

nodes and vascular compromise by neoplastic destruction of pancreatic vessels.[8] In the present case, obstruction of the pancreatic duct by diffuse metastasis in the duodenal wall could have caused acute pancreatitis.

Diagnosis of GI metastasis from breast cancer is sometimes difficult prior to surgical intervention, and primary GI carcinoma may mistakenly be diagnosed.[6,7] In GI metastases, the infiltrating tumour cells are located in the submucosal layer or deeper with minimal mucosal lesions.[6,7] Because findings of diagnostic imaging are frequently not specific for GI metastasis, GI endoscopy with deep biopsy is recommended for accurate diagnosis.[6] In the present case, although neither physical nor ultrasound examination detected abnormalities in the breasts, breast biopsy was performed due to abnormal enhancement on CT in the affected breast, and histopathological and immunohistochemical comparison of findings enabled the diagnosis of breast SRCC with duodenal metastasis. Radiological modalities including CT and ultrasound may not detect ILC.[9] It is thus important to be aware of breast cancer as a possible origin of GI metastases.

For GI metastasis of breast cancer, systemic treatment is the main form of primary therapy, since this condition is frequently associated with widespread metastases. Surgical intervention does not extend patient survival, but can be considered in patients with bowel obstruction or bleeding.[6] In our case, despite the presence of widespread metastases, multiple palliative surgeries were performed for duodenal and large intestinal obstruction, resulting in maintenance of oral intake without bowel obstruction. Because cytotoxic chemotherapy is not always a promising mode of treatment, surgical intervention may, in selected patients, yield a better outcome than chemotherapy alone.[10]

References

1. Asch MJ, Wiedel PD, Habif DV. Gastrointestinal metastases from carcinoma of the breast: autopsy study and 18 cases requiring operative intervention. *Arch Surg* 1968;96: 840–3.
2. Kondo Y, Akita T, Sugano I, Isono K. Signet ring cell carcinoma of the breast. *Acta Pathol Jpn* 1984;34:875–80.

3. Kobayashi T, Shibata K, Matsuda Y, et al. A case of invasive lobular carcinoma of the breast first manifesting with duodenal obstruction. *Breast Cancer* 2004;11:306–8.
4. Hull MT, Seo IS, Battersby JS, Csicsko JF. Signet-ring cell carcinoma of the breast: a clinicopathologic study of 24 cases. *Am J Clin Pathol* 1980;73:31–5.
5. Ashikari R, Huvos AG, Urban JA, Robbins GF. Infiltrating lobular carcinoma of the breast. *Cancer* 1973;31:110–6.
6. Schwarz RE, Klimstra DS, Turnbull ADM. Metastatic breast cancer masquerading as gastrointestinal primary. *Am J Gastroenterol* 1998;93:111–4.
7. Merino MJ, Livolsi VA. Signet ring carcinoma of the female breast: a clinicopathologic analysis of 24 cases. *Cancer* 1981;48: 1830–7.
8. Hung SSJ, Ardalan B. Metastasis-induced acute pancreatitis. *West J Med* 1991;155:176–8.
9. Uchiyama N, Miyakawa K, Moriyama N, Kumazaki T. Radiographic features of invasive lobular carcinoma of the breast. *Radiat Med* 2001;19:19–25.
10. Pappo I, Feigin E, Uziely B, Amir G. Biliary and pancreatic metastases of breast carcinoma: is surgical palliation indicated? *J Surg Oncol* 1991;46:211–4.