

• Case Report •

## Carcinoma of the right side colon accompanied by Sister Mary Joseph's nodule and inguinal nodal metastases: a case report and literature review

Yong-You Wu<sup>1</sup>, Chun-Gen Xing<sup>1</sup>, Ju-Xiang Jiang<sup>2</sup>, Xiao-Dong Lu<sup>1</sup>, Yi-Zhong Feng<sup>3</sup>, Hao-Rong Wu<sup>1</sup>

<sup>1</sup> Department of General Surgery, the Second Affiliated Hospital of Soochow University, Suzhou, Jiangsu 215004, P.R. China; <sup>2</sup> Department of Biochemistry and Molecular Biology, Soochow University, Suzhou, Jiangsu 215000, P.R. China; <sup>3</sup> Department of Pathology, the Second Affiliated Hospital of Soochow University, Suzhou, Jiangsu 215004, P.R. China

**[Abstract]** Umbilical metastases from intraperitoneal malignancies are universally referred to Sister Mary Joseph's nodule (SMJN). The most frequent primary tumor sites include the stomach and ovaries. SMJN caused by colon cancer is uncommon. Likewise, carcinoma of the right side colon metastasizing to inguinal lymph nodes is considered almost impossible. To the best of our knowledge, there is no report of right side colon cancer synchronously involving both the umbilicus and inguinal lymph nodes in the literature. We present a case of right side colon cancer (RSCC) metastasizing to the umbilicus and inguinal lymph nodes, which was confirmed by routine pathological evaluation and immunohistochemistry.

**Key words:** Colon neoplasm, Sister Mary Joseph's nodule, umbilical metastasis, inguinal lymph node

### Case description

A 37-year-old man was admitted to the Second Affiliated Hospital of Soochow University for intermittent abdominal pain and bloody stools for one year. Three months before, enlargement of pre-existing inguinal lymph nodes with mild dull pain was noted in bilateral inguinal regions. Two weeks before, a painless mass in the umbilicus was accidentally palpated. Physical examination revealed an umbilical mass, 2 cm in diameter, with slightly reddish skin. The mass was hard and fixed to the abdominal wall. Multiple lymph nodes with mild tenderness were palpable in bilateral inguinal regions. The nodes were confluent and slightly movable with a maximal diameter of 2.5 cm. Under colonoscope, a proliferative lesion in the hepatic flexure of the colon was discovered and further observation of the proximal colon was hampered by luminal stricture caused by the lesion. On CT scans, mural thickening of the ascending colon and cecum, intumescent intra-abdominal, retroperitoneal and bilateral inguinal lymph nodes, filling defect in the hepatic flexure and cystic lesions of the liver, and an enhanced umbilical mass with obscure border were observed (Figure 1). The level of serum

carcinoembryonic antigen (CEA) was elevated (55 ng/mL). This case was diagnosed preoperatively as colon carcinoma with umbilical metastases, and inguinal nodal metastases was suspected. Due to severe intestinal spasm and bloody stool, the patient insisted on surgical treatment, though systematic chemotherapy was suggested. Right hemicolectomy and resection of the umbilical mass were performed through a median abdominal incision. Excision of the enlarged lymph nodes of the right groin was performed for biopsy. Abdominal exploration revealed miliary dissemination in the Douglas pouch and cancerous infiltration in the right diaphragm. No obvious metastatic loci were detected on the surface of the liver. Pathologic examination confirmed colon adenocarcinoma as well as umbilical metastasis and inguinal lymph node metastasis (Figure 2). Immunohistochemical analysis with anti-CEA and anti-CK20 antibodies revealed positive staining of tumor cells from the above three sites (Figure 3). This case was diagnosed as stage IV disease and chemotherapy was recommended. However, the patient refused further therapy and died four months after operation.

### Discussion

#### Umbilical metastasis from colon cancer

Cutaneous metastases to the umbilicus are universally referred to as Sister Joseph's (or Sister Mary Joseph's) nodule (SMJN)<sup>1,2</sup>. Though uncommon, SMJN is well documented and generally regarded as a unfavorable prognostic sign with survival ranging from 2 to 11 months from the time of initial diagnosis in

Correspondence to: Ju-Xiang Jiang; Tel: +86-512-65880108; Tel: +86-512-68284303; Email: jiangjuxiang@suda.edu.cn

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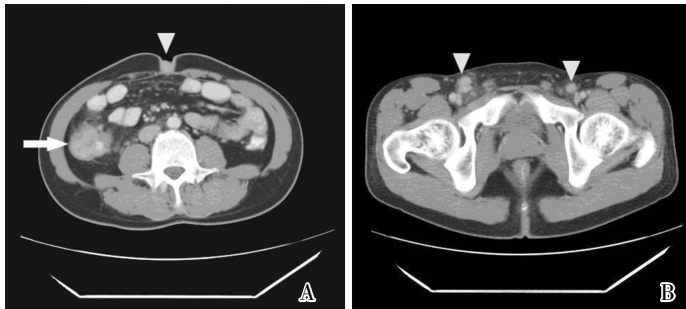


Figure 1 CT scans of the abdomen and inguinal regions of a 37-year-old man with right colon cancer accompanied by Sister Mary Joseph's nodule and inguinal nodal metastases

A, Contrast-enhanced CT scan shows a large, circumferential soft tissue mass in the ascending colon and an umbilical mass. Luminal narrowing, irregular wall thickening can also be noted (arrow). The umbilical mass shows enhancement with slightly obscure contour. Peritoneal involvement is suspected (arrow head). B, Contrast-enhanced CT scan of the inguinal region shows bilateral enlarged inguinal lymph nodes with enhancement and obscure contour, suggesting inguinal metastases.

untreated patients<sup>1-3</sup>. The primary lesions are mostly discovered in the stomach and ovary, whereas colon cancer seldom spreads to the umbilicus. The first report of umbilical metastasis from colon cancer was published in 1846 by Walshe. In a review by Shetty<sup>4</sup>, 265 cases of metastatic tumors of the umbilicus were reported from 1830 to 1989, among which 17 cases were found to be originated from colon cancer. Galvan<sup>5</sup> summarized the characteristics of 407 umbilical metastases from 1966 to 1997 and found that 14.6% of all cases were originated from colorectal cancers. Dodiuk-Gad *et al.*<sup>6</sup> reported another case of SMJN caused by colon cancer. As to SMJN originating from the right side of colon cancer, there are only case reports. According to Gabriele *et al.*<sup>7</sup>, SMJN originated from cecum cancer is very rare and only four cases have been reported in literature, though Moll<sup>8</sup> reported another case in 1996.

The underlying mechanisms of SMJN remain unclear with several hypotheses on the spread patterns of this special cutaneous metastasis. Contiguous, hematogenous, lymphatic spread and direct extension along the ligaments of embryonic

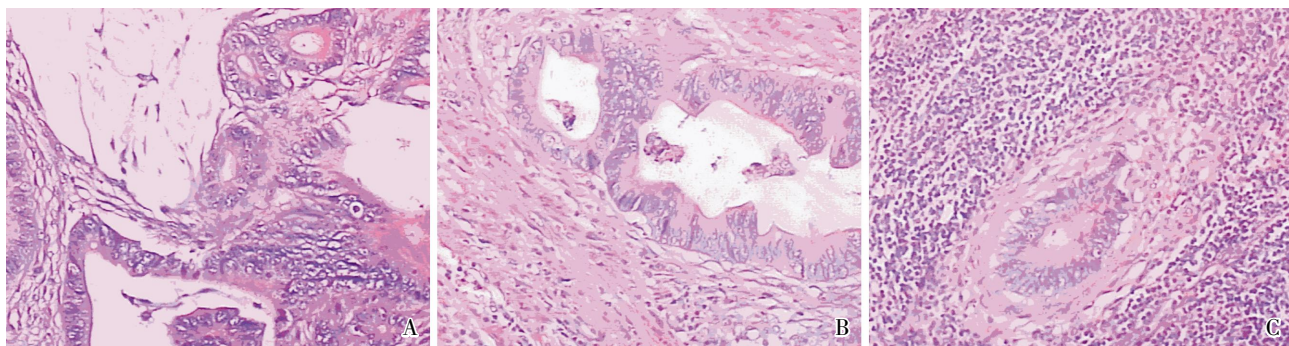


Figure 2 Morphology of the primary cancer and metastatic specimens (HE ×100)

A, The primary colon adenocarcinoma shows tubular structures. B, The Sister Mary Joseph's nodule shows stained collagen and similar glandular structures with primary lesion. C, The inguinal lymph node shows nodal structures and similar structures of primary lesion.

origin are the metastasis modes of SMJN<sup>1,2,7,9</sup>. In our case, the peritoneal involvement underlying SMJN was confirmed by both CT and pathology, indicating the possibility of contiguous spread from intra-abdominal malignancy. Consistent with previous reports, the SMJN in our patient lacked typical structures of lymph nodes, excluding the possibility of typical lymphatic spread. Due to the common association between hepatic and umbilical metastases, it is hypothesized that gastroenterological malignancies may spread to the liver through the portal system and then to the umbilicus through lymphatic or venous channels. Though hepatic spread is noticed in most patients with SMJN, liver involvement does not seem to be a prerequisite for this cutaneous metastasis. Though obvious metastasis of the liver was not noted in our patient, spread through the portal system could not be safely excluded from consideration.

### Inguinal metastasis from colon cancer

The most common sites of distant metastasis of colon carcinoma are the liver, lungs and bones. Though metastases to other unusual sites, such as the breast and thyroid glands, have

been sporadically reported in literature, colon carcinoma, especially that of the right half colon, spreading to inguinal lymph nodes is considered almost impossible due to the anatomical features and cranially directed lymphatic drain of the colon. Inguinal metastases from rectal cancer can be presumed due to the blockade of ascending lymphatic flow by proximal metastatic lymph nodes, interruption of lymphatic drain by previous lymphadenectomy or pelvic recurrence<sup>10</sup>. The same theory can not be reasonably applied to inguinal spread of the right side colon cancer. Though several cases of such metastases have been reported in the Chinese and Japanese literature, no such reports have been found in the English literature by searching through PubMed. Morton *et al.*<sup>11</sup> reported an unusual case of ascending colon cancer metastasizing to the right inguinal region, but it was a subcutaneous metastasis, not lymph node metastasis.

### Concurrent umbilical and inguinal metastases from colon cancer

To the best of our knowledge, no report of concurrent

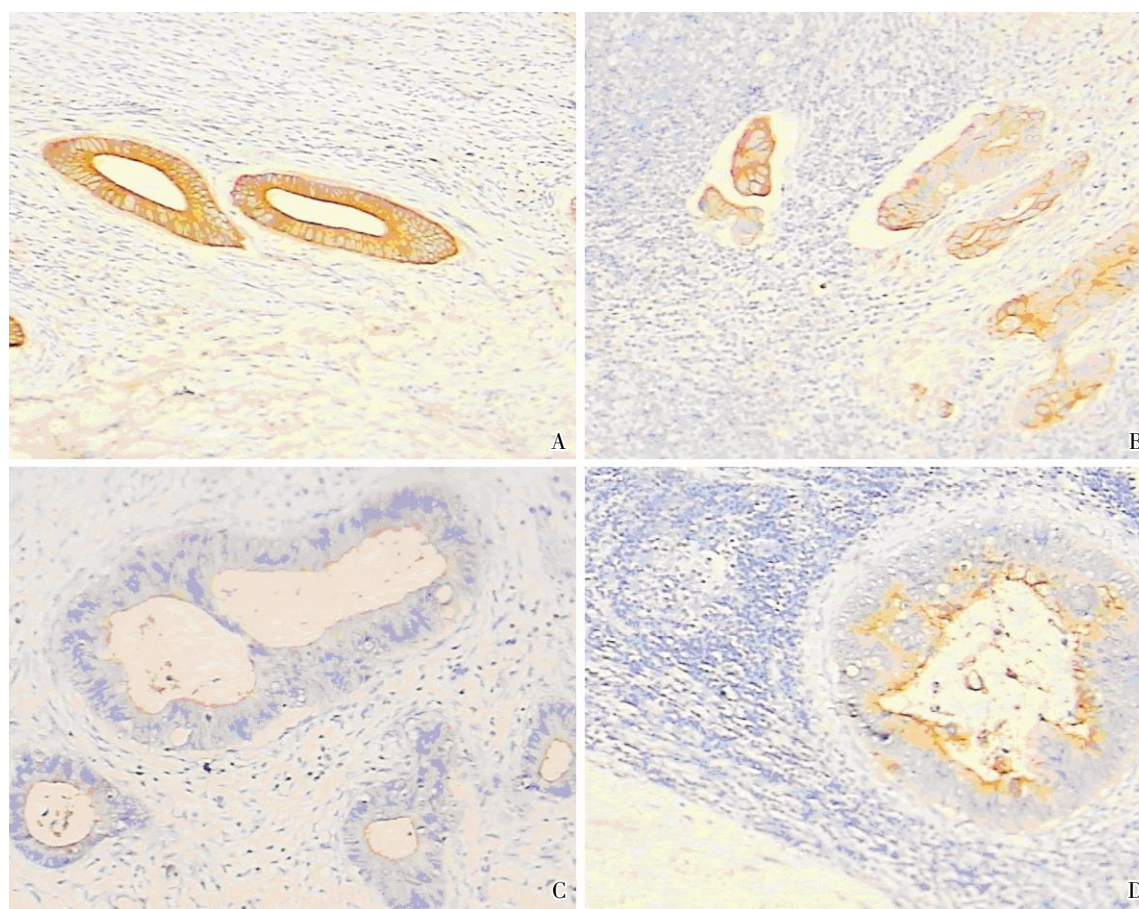


Figure 3 Immunohistochemical staining of cancer cells in metastatic lesions (×100)

A, Positive staining of umbilical metastasis with anti-CK20 antibody. B, Positive staining of inguinal lymph nodes with anti-CK20 antibody. C, Positive staining of umbilical metastasis with anti-CEA antibody. D, Positive staining of an inguinal lymph node with anti-CEA antibody.

umbilical and inguinal metastases from colon cancer is available in the literature. In the present case, although intumescent inguinal lymph nodes were found prior to the umbilical mass, the sequence of progression remains obscure. One reasonable hypothesis may be that inguinal lymph node metastasis occurred secondary to the umbilical metastasis via the lymphatic pathway of the abdominal wall. Yet, SMJN and inguinal lymph node metastasis may be separate events. Due to the intra-abdominal dissemination of the primary lesion, tumor spreading along the iliac vessels to the inguinal lymph nodes is also a possible route of metastasis.

The patient noticed the enlargement and discomfort of the pre-existing inguinal lymph nodes. Bilateral metastases to pre-existing inguinal lymph nodes demonstrate that diseased lymph nodes are prone to arrest and harbor circulating cancer cells and develop into metastatic loci.

The value of surgery for SMJN is still controversial<sup>1-3,7</sup>. Due to the devastating prognosis of those patients, conservative treatment and best supportive care are recommended by most researchers. However, no report on the management of colon cancer patients with SMJN is available. During the last few years, great progress has been made on chemotherapy and targeted

therapy for colon cancer. Significantly prolonged median survival time and improved quality of life have been reported<sup>12-14</sup>. Complicated with inguinal metastasis or not, SMJN originating from colon cancer is an ominous sign of prognosis. Due to the limited role of operation and remarkable advances in chemotherapy and targeted therapy, surgical intervention should not be recommended for patients without severe complications, such as bleeding, obstruction and perforation.

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