Leiomyomatosis of Mesenteric Lymph Nodes Associated with Duodenal Adenocarcinoma

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Leiomyomatosis of lymph nodes is an extremely rare disease. Only a few cases have previously been reported in pelvic lymph nodes. They were related to a benign uterine leiomyoma, a metastasizing uterine leiomyoma, an endometrial adenocarcinoma, and an ovarian endometrioid carcinoma. We report on a case of leiomyomatosis of the mesenteric lymph nodes associated with a duodenal adenocarcinoma with no history of uterine leiomyoma or any gynecological malignancy. The patient, a 56-year-old woman, was found to have an adenocarcinoma of the duodenum. All mesenteric lymph nodes removed showed leiomyomatosis, which was verified by immunohistochemical study showing positive immunostaining for smooth muscle actin, desmin, and vimentin, but negative staining for HMB-45. It is necessary to make a differential diagnosis from other examples of spindle cell proliferation involving lymph nodes such as a hemorrhagic spindle cell tumor with amianthoid fibers (palisade myofibroblastoma), angiomyolipoma, lymphangiomyomatosis, inflammatory pseudotumor, and Kaposi's sarcoma. *(Chang Gung Med J 2002;25:271-4)*

Key words: leiomyomatosis, mesenteric lymph node, duodenal adenocarcinoma.

Leiomyomatosis of the lymph nodes has been previously reported in association with a benign uterine leiomyoma[(1)] or related to the so-called uterine benign metastasizing leiomyoma,[(2)] disseminated peritoneal leiomyomatosis,[(1)] leiomyomatosis in pelvic lymph nodes without a uterine leiomyoma, and disseminated peritoneal leiomyomatosis.[(3)]

We report on a case of leiomyomatosis of the mesenteric lymph nodes associated with a duodenal adenocarcinoma. To our knowledge, this association has not been reported previously.

CASE REPORT

A 56-year-old postmenopausal woman with no history of uterine leiomyoma, endometriosis, or any gynecological malignancy was admitted to Chang Gung Memorial Hospital in July 1996 because of postprandial vomiting and abdominal distension. All laboratory data were within normal limits. An upper gastrointestinal radiographic study revealed an intraluminal filling defect in the duodenojejunal junction, and a computed tomographic scan showed gastric outlet obstruction and regional lymph nodes with increased enhancement (Fig. 1). Under the impression of a duodenal tumor, segmental resection of the duodenum with end-to-side anastomosis and mesenteric lymph node dissection were performed.

Histological examination showed a moderately differentiated adenocarcinoma of the duodenum. All 10 dissected mesenteric lymph nodes were replaced by fascicles of spindle cells with bland-looking, mitotically inactive cells (Figs. 2, 3). Immunohistochemical study showed a strongly positive reaction for smooth muscle actin (Fig. 4), desmin, and vimentin, but it was negative for HMB-45.

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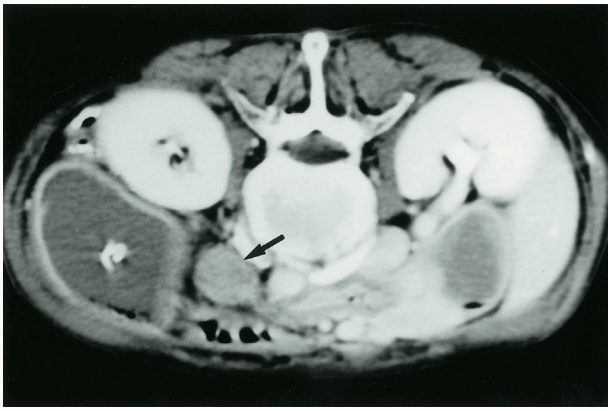

Fig. 1 Enhanced computed tomogram showing duodenal obstruction with an enlarged paragastric lymph node (arrow).

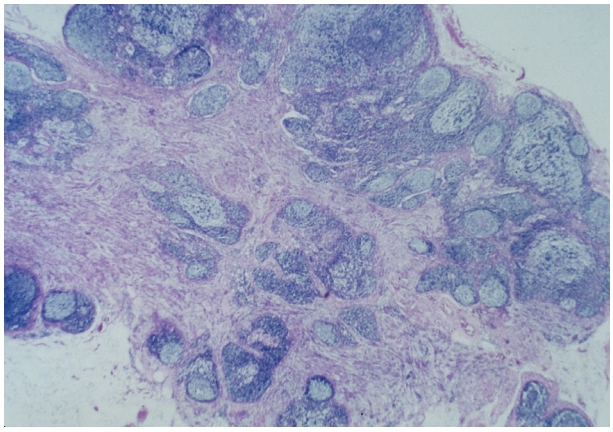

Fig. 2 Extensive replacement of the lymph node by spindle cell proliferation. (H & E stain, 40¡)

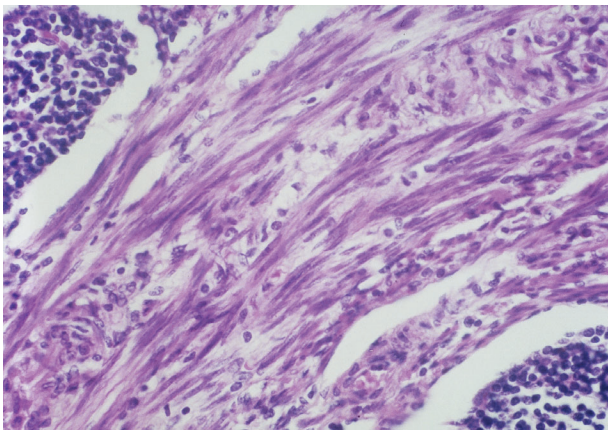

Fig. 3 Bland-looking spindle cells which lack mitoses. (H & E stain, 200¡)

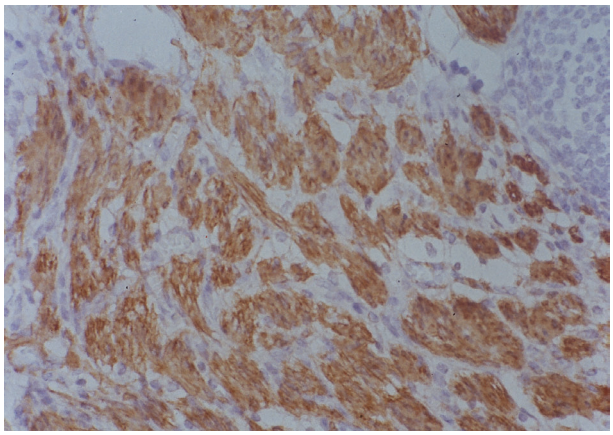

Fig. 4 Immunohistochemical staining showing strongly positive reaction for smooth muscle actin. (200¡)

Therefore, a diagnosis of leiomyomatosis of the lymph nodes was made.

DISCUSSION

Leiomyomatosis of the lymph nodes is very rare. Leiomyomatosis of pelvic lymph nodes has been reported in association with a benign uterine leiomyoma[1] or a metastasizing uterine leiomyoma.[2] The pathogenesis was postulated to have resulted from uterine tumor cells spontaneously gaining access to lymphatic channels or during manipulation at uterine curettement.[4,5] It was also reported to be associated with an endometrial adenocarcinoma[6] and an ovarian endometrioid carcinoma.[7] The association with a duodenal adenocarcinoma has not been reported before.

Leiomyomatosis involving lymph nodes must be distinguished from other spindle cell tumors involving the lymph nodes, such as a hemorrhagic spindle cell tumor with amianthoid fibers (palisade myofibroblastoma),[8] an angiomyolipoma,[9] lymphangiomyomatosis,[9] an inflammatory pseudotumor,[10] and Kaposi's sarcoma.[11] The hemorrhagic

spindle cell tumor with amianthoid fibers is accompanied by a hemorrhagic rim and the formation of amianthoid fibers.[8] The spindle cells are immunoreactive for vimentin, muscle-specific actin, and myosin, but not desmin.[8] Lymphangiomyomatosis and angiomyolipomas are positive for HMB-45, muscle-specific actin, and desmin.[9] But leiomyomatosis involving lymph nodes will positively immunostain for smooth muscle actin, desmin, vimentin, but negatively for HMB-45. An inflammatory pseudotumor of the lymph nodes shows proliferation of spindle cells, blood vessels lined by flat endothelium, and inflammatory cell infiltrate; it differs from leiomyomatosis of the lymph nodes because of its inflammatory character, being particularly rich in plasma cells.[10] Kaposi's sarcoma of the lymph nodes is characterized histologically by an admixture of proliferating spindle cells separated by slit-like spaces with extravasated red blood cells in the lymph node.[11]

The present case is interesting because of the multiple lymph node involvement. It could be a reactive change of nearly neoplasm rather than true neoplastic change. It is believed that leiomyomatosis of the lymph nodes arises primarily from the smooth muscle elements of the capsule or vascular wall and is presumably caused by the so-called myogenic factor that drains into regional lymph nodes from a host tumor.

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十二指腸腺癌併腸繫膜淋巴腺平滑肌變化

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淋巴腺之平滑肌變化是一種極爲罕見的疾病，文獻中祇有幾例在骨盆淋巴腺中出現此種變化曾被報導過。這些變化過去認爲和良性子宮肌肉瘤、轉移性子宮肌肉瘤、子宮內膜腺癌和卵巢之子宮內膜樣癌有關。本報告的病例是十二指腸腺癌的病人，在腸繫膜淋巴腺出現平滑肌變化，但之前沒有子宮平滑肌內瘤或任何婦科惡性腫瘤的病史。病人是五十六歲的女性，在十二指腸處有腺癌。所有摘除的腸繫膜淋巴腺，經由免疫螢光研究呈現平滑肌肌動蛋白陽性、硬纖維素陽性、間質纖維陽性、HMB-45陰性，顯示其具有平滑肌變化。此變化需和一些波及淋巴腺的梭狀細胞增生疾病作鑑別診斷，例如：出血性併石綿纖維之梭狀細胞腫瘤、血管肌肉脂肪瘤、淋巴血管平滑肌變化、發炎性僞瘤和卡波西氏肉瘤等。(長庚醫誌 2002;25:271-4)

關鍵字：平滑肌變化，腸繫膜淋巴腺，十二指腸腺癌。

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