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[Case Report]

GASTROINTESTINAL STROMAL TUMOR MIMICKING GYNECOLOGICAL DISEASE

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Abstract: Gastrointestinal stromal tumors (GISTs) are common mesenchymal tumors of the digestive tract. These tumors occasionally present a pelvic mass and leading to the misdiagnosis of gynecologic diseases. Two patients with GIST in the small intestine giving an impression of an ovarian fibroma and a uterine leiomyoma respectively were diagnosed correctly at the surgery. In the patients with a pelvic mass, especially if unusual symptoms and laboratory data being not compatible with gynecological disease, the possibility of diseases other than a gynecologic disease has to be considered.

Key words: Gastrointestinal stromal tumor, preoperative diagnosis, ovarian fibroma, uterine leiomyoma

INTRODUCTION

GISTs are mesenchymal tumors of the digestive tract[1-3)]. These tumors differentiate toward a pace maker cell phenotype and express a KIT tyrosine kinase receptor[4,5)]. For the reason of the mesencymal nature of tumor, GISTs are composed of spindle cells and referred to as leiomyoma or leiomyosarcoma histologically. Because of these histological characters, if GISTs present a pelvic tumor, they occasionally lead to us make misdiagonsis of gynecologic diseases, such as uterine leiomyomas and ovarian tumors. In this report, we described two cases with GISTs

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mimicking gynecological diseases. Furthermore, we discussed the cause of misdiagnosis.

CASE REPORT

Case 1

A 69-year-old Japanese woman, 3 para, 3 gravid was admitted to our hospital with an abdominal pain as the chief complaint on October 2, 2003. She had received total hysterectomy and left oohrectomy at age of 49 for a uterine leiomyoma. Physical examination showed a fist-sized, elastic-firm, movable mass in the pelvis. Computed tomography (CT) revealed a multinodular, solid tumor with partially

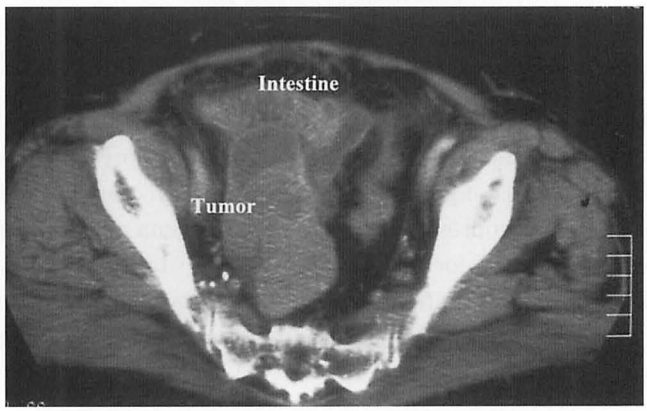

Fig. 1. CT of case 1 revealed a pelvic tumor, 7×4 cm in size. Tumor was composed of solid, homogenous part and foci of cystic lesion. The small intestine was adjacent to the tumor.

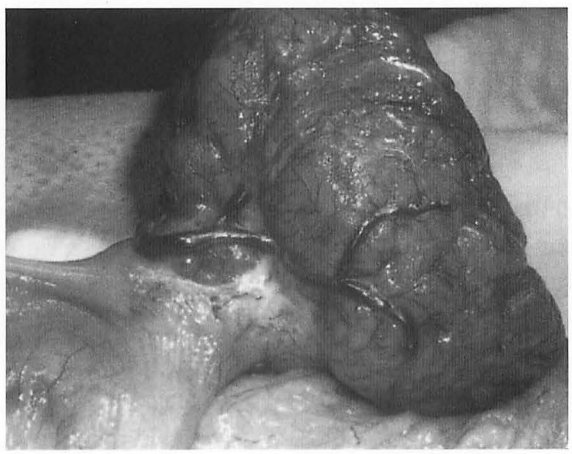

Fig. 2. Surgical finding of case 1 showed the tumor arising from the ileum 40 cm proximal from ileum end, grew extraluminally and measured 10×6×6 cm.

cystic lesion in the pelvis. The small intestine was adjacent to the tumor (Fig. 1). Ultrasoundgraphy revealed pelvic tumor with homogenous echogenicity. For the preoperative diagnosis of ovarian theco-fibroma, the operation was performed on Octorber 20, 2003. At the surgery, a tumor revealed a subserosal tumor 10×6 cm in size, arising from the ileum, 40 cm oral-sided from ileum-end and located in the pelvic cavity (Fig. 2). The atrophic right ovary was normal in appearance. Partial resection of the ileum was performed. Surgical findings suggested a GIST of small intestine and a GIST was defined by the pathological examination. The patient received no adjuvant therapy and is free from disease 13 months after diagnosis.

Case 2

A 39-year-old Japanese female, 2 para, 2 gravida visited our hospital for general fatigue on December 19, 2003. Laboratory data revealed anemia (Hb 6.3 g/dl). A fist-sized, elastic firm tumor was palpated during the pelvic examination. Her stool was positive for occult blood. Ultrasoundgraphy and CT revealed a solid mass neighboring the uterus (Fig. 3 & 4). The echogenicity and density of the tumor was homogenous. Under preoperative diagnosis as an uterine subserosal leiomyoma, she underwent an operation on December 4, 2003. At the surgery, a tumor revealed a subserosal tumor 8×7 cm in size, arising from the ileum, 25 cm oral-sided from the ileum-end and located in the pelvic cavity (Fig. 5). The uterus and bilateral ovaries were normal in appearance. Partial resection of the ileum was performed. Surgical findings suggested a GIST of small intestine and a GIST was defined by the pathological examination. The patient received no adjuvant therapy and is free from disease 12 months after diagnosis.

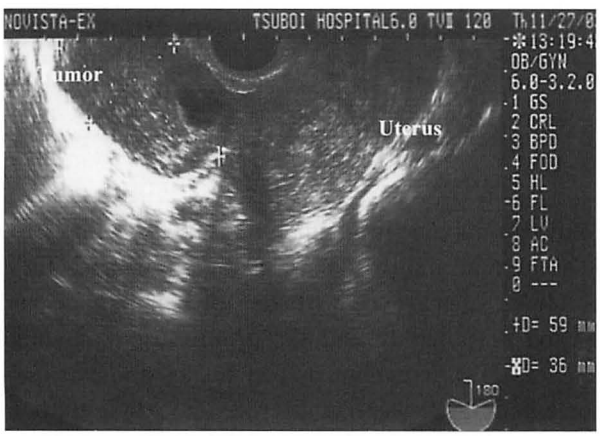

Fig. 3. Ultrasoundgraphy of case 2 revealed 6×3.5 cm-sized tumor, neighboring the uterus.

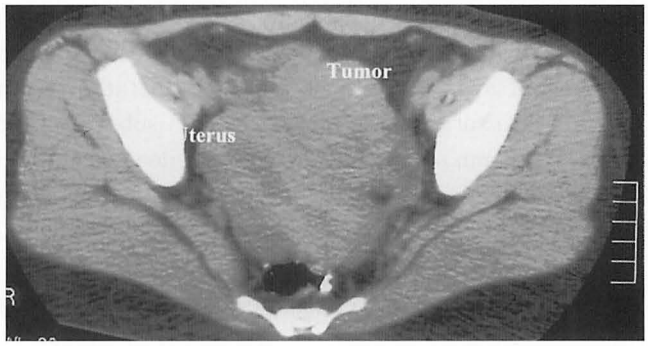

Fig. 4. CT of case 2 revealed pelvic mass, 8×5 cm in size. The tumor being adjacent to the uterus gave an impression of uterine leiomyoma.

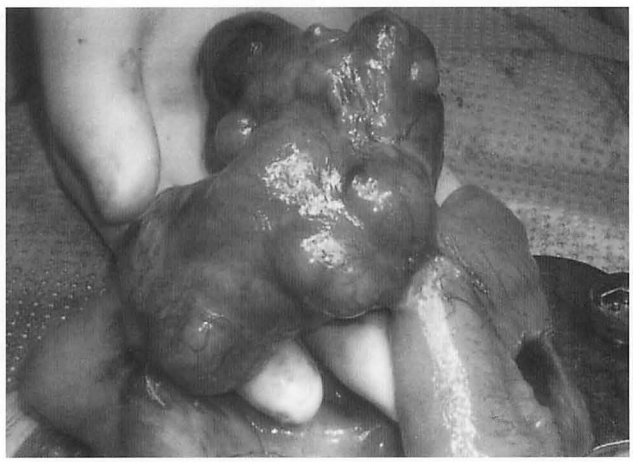

Fig. 5. Surgical finding of case 2 showed the multinodular, elastic firm tumor arising from the ileum and measured 8×6×6 cm.

PATHOLOGICAL FINDINGS

Case 1

A tumor revealed a submucosal tumor and extraluminal growth from ileum. The tumor was solid and multinodular. On cut surface, the tumor revealed homogenous tan-colored appearance with foci of hemorrhage and necrosis. The tumor was composed of a highly cellular proliferation of spindle cells (Fig. 6). Cytologic atypia containing irregular-shaped nuclei and various-sized cells was evident. Multinucleated cells were scattered. Focally, epithelioid components were found. Tumor cells arranged in glandular clusters surrounded slit like vasucular cannals (Fig. 7). The nuclei of tumor cells revealed hyperchromatic and showed coarse chromatin and predominant nucleoli. The mitotic figure count was 3 per 10 high power fields. Theses histological findings suggested a malignant

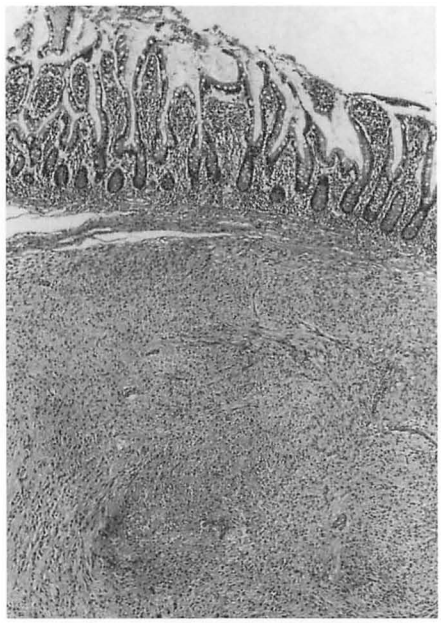

Fig. 6. Histological specimen reveals the well- circumscribed tumor locating in submucosal layer. (H & E stain, ×40)

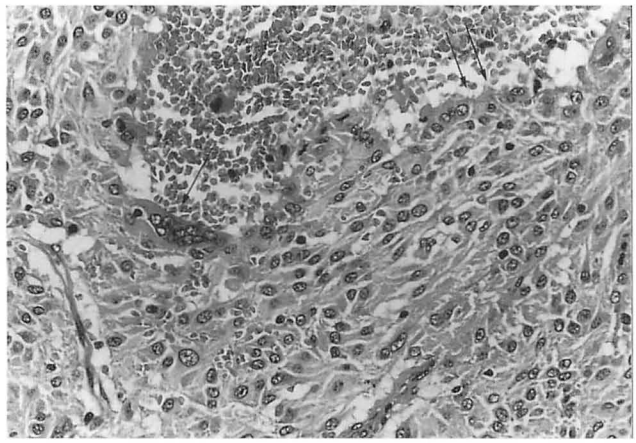

Fig. 7. In specimen of case 1, the tumor is composed of high cellular proliferation of spindle cells. Vague epithelioid arrangement of tumor cells and multinucleated giant cells are scatteredly observed. (Arrows) (H & E stain, ×300)

potential of this tumor. Tumor cells were positive for c-kit and CD34 immunohistochemically (Fig. 8).

Case 2

A tumor revealed a submucosal tumor accompanied with ulceration of superficial intestinal mucosa. The tumor was solid and showed extraluminal

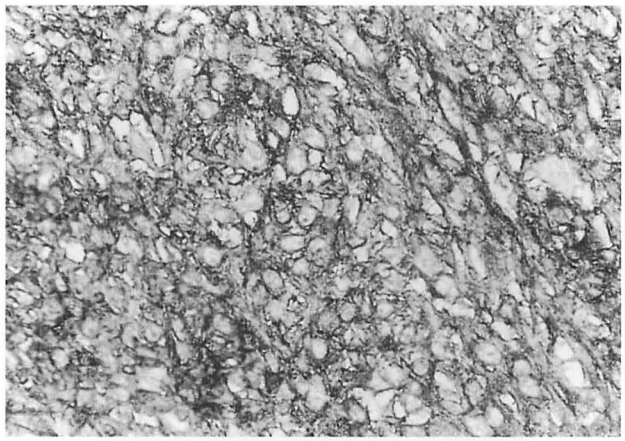

Fig. 8. Immunohistochemical staining of cases 1 reveals diffusely positive reaction for c-kit in cellular membrane of tumor cells. (ABC stain, ×600)

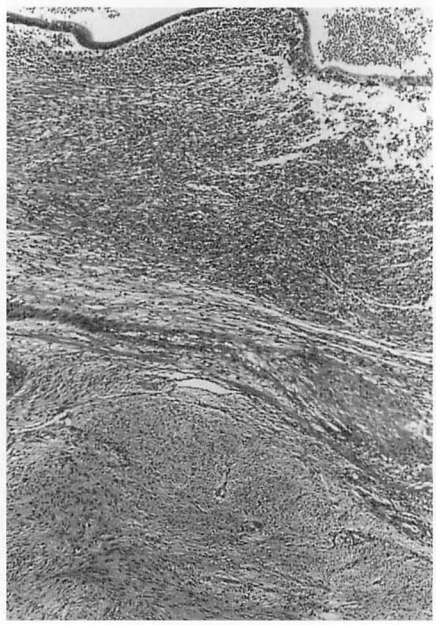

Fig. 9. Specimen of case 2 reveals well- circumscribed, submucosal tumor composed of uniform, spindle-shaped cells. The overlying intestinal mucosa of the tumor is associated with ulceration and shows regeneration of epithelium. (H & E stain, ×40)

growth. On cut surface, the tumor revealed homogenous tan-colored appearance. The tumor was composed of a uniform population of spindle cells with an abundant eosinophilic cytoplasm. The intestinal mucosa overling the tumor revealed ulceration and regeneration of epithelium (Fig. 9). Tumor cells were divided into nests by a fine vascular septum and the cellularity was low. Tumor cells were uniform and

had a fine nuclear chromatin. The mitotic figure was not evident. The tumor fulfilled the criteria for benign tumors. Tumor cells were positive for c-kit and CD34 immunohistochemically.

DISCUSSION

This report presents two cases of GISTs, one mimicking ovarian theco-fibroma and the other a uterine leiomyoma. GISTs are an entity of most commonly mesenchymal neoplasm in the digestive tract and composed of spindle or epithelioid cells[1-3]. These tumors are evaluated in a specific manner, clinically and pathologically. GISTs have been found to differentiate toward the phenotype of interstitial cells of Cajal as the pace maker cells and expressed kit tyrosine kinase[4,5]. The histological and immunohistochemical findings in our two cases were compatible with GISTs[6]. These tumors predominantly arise in the middle-aged and elderly people. Approximately one third of GISTs arose in the small intestine[7]. In the past, GISTs revealing spindle cell tumors have often been referred to as leiomyoma or leiomyosarcoma of the digestive tract, based on their purported morphologic resemblance to smooth muscle tumors in other anatomic sites[8]. Reflecting these histopathological characteristics, GISTs occasionally may present well-movable, multinodular tumors with a smooth surface[8]. Images of most GISTs may show a well-circumscribed border and homogenous density[9]. If tumors exist in the pelvis, these findings give the impression of gynecologic disease such as leiomyomas of uterus or fibrous tumors of ovary. In such circumstances they are first noticed by gynecologists and are often diagnosed, prior to surgery, as gynecological disease. Several gynecologists reported GISTs mimicking gynecological diseases. Belics et al. have reported a patient with GIST representing a large pelvic mass[10]. The tumor gave an impression of an ovarian tumor. Carlomagno et al. have reported a 42-year-old woman with GIST masquerading as an ovarian cancer[11]. Zighelboim et al. have reported a 31-year-old woman with GIST presenting a pelvic mass[12].

Ovarian theco-fibroma is rare but not so in the middle and old aged women. Theco-fibromas reveal an elastic firm, multinodular tumor with a smooth surface. Images of theco-fibroma are solid, showing uniform density. Our first case with a pelvic tumor gave an impression of ovarian fibroma. However her complaint of abdominal pain and tumor neighboring the small intestine in CT suggested a digestive tract disease. Leiomyoma of the uterus is the most common causation of anemia in the young-middle aged female. In our second case, patients with a pelvic mass and anemia gave an impression of uterine leiomyoma. Her tumor neighboring the uterus having multinodular, uniform appearance mimicked uterine leiomyoma. However her stool was positive for occult blood and tumor associated ulceration. So a disease of the digestive tract should be considered.

In the patients with pelvic masses, especially if unusual signs and clinical data

(for example colic abdominal pain, positive occult blood in stool) were present, the possibility of a non-gynecologic tumor must be considered and the support of a digestive tract surgeon should be enlisted for treatment.

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