CT findings of sclerosing stromal tumor of the ovary: A report of two cases and review of the literature

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Received January 16, 2015; Accepted February 16, 2016

DOI: 10.3892/ol.2016.4441

Abstract. Sclerosing stromal tumor (SST) of the ovary, which was first described by Chalvardjian and Scully in 1973, is a rare ovarian neoplasm, occurring predominantly in young women. The most common clinical symptom in patients with SST is menstrual irregularities. Microscopically, the tumor is characterized by the presence of pseudo-lobulated cellular areas, with a prominent tendency to sclerosis, marked vascularity and pronounced variation in cellular size and shape. In the current study, 2 cases of SST of the ovary are presented. These cases were confirmed by imaging, surgical and histological examination. No adjuvant therapy was administered to the patients and the two patients were disease-free with no imaging findings of recurrence or metastasis 24 months following surgery.

Introduction

Sclerosing stromal tumor (SST) of the ovary is a rare benign tumor that has been classified as a sex cord-stromal tumor and is predominantly observed in young women (1,2). SSTs account for 1.5% of all ovarian tumors (1). The tumor usually occurs in the second and third decades of life (mean age, 21 years). The most common presenting symptoms are menstrual irregularity, abdominal pain and a lower abdominal mass (1,2). Microscopically, the tumor is characterized by pseudo-lobulation of cellular areas, marked vascularity and significant variation in cellular size and shape, and sclerosis is common (1,3). Surgical removal is curative and no local or distant recurrences have been reported following surgery (2,3). To the best of our knowledge, there have been numerous reports regarding the microscopic, ultrastructural and immunohistochemical, but not imaging, findings associated with this tumor (3). The present study describes the computed tomography (CT) findings of 2 patients with ovarian SST, and confirms the correlation between imaging and pathological findings, as well as the reliability of imaging in preoperative characterization. The study was approved by the Ethics Committee of Subei People's Hospital of Jiangsu Province (Yangzhou, China) and the patients provided written informed consent for the publication of the present study.

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Key words: ovary, sclerosing stromal tumor, computed tomography

Case report

Case 1. An 18-year-old Chinese female patient was admitted to Subei People's Hospital of Jiangsu Province in July 2012 with menstrual irregularities for 6 months. The blood test results for blood cell count, biochemistry and tumor markers, including cancer antigen (CA)19-9 (3.57 KU/l; normal range, <35.00 KU/l), carcinoembryonic antigen CA125 (5.15 KU/l; normal range, <35.00 KU/l) and α-fetoprotein (3.57 ng/ml; normal range, <20.00 ng/ml) were all normal. The levels of serum hormones, including estradiol (60 ng/ml; normal range, 22-144 ng/ml), progesterone (8.13 ng/ml; normal range, 5.16-18.56 ng/ml) and testosterone (12.3 ng/ml; normal range, 10.0-75.0 ng/ml), were also normal.

Non-enhanced CT (Lightspeed VCT 64; GE Healthcare, Milwaukee, WI, USA) scan revealed a round soft tissue mass with clear boundaries in the middle of the pelvis. The mass measured 76x77 mm and displayed non-homogeneous density, with solid, non-calcified tissue at the periphery and a fluid, patch-shaped area in the inner zone (Fig. 1A). Plain CT values were 31-45 HU, with a mean value of 37.6 HU. Intravenous bolus injection of iodinated contrast medium (iopromide; Ultravist® 320; Bayer HealthCare Pharmaceuticals, Berlin, Germany) yielded early nodular and ring enhancement of the peripheral portion of the mass, which was as high as that of the vessels in the arterial phase (Fig. 1B). In the venous phase, the degree of enhancement was decreased, but the area of enhancement increased with centripetal progression (Fig. 1C). The CT values in the arterial and venous phases were 145-150 HU, with a mean value of 147 HU, and 128-135 HU, with a mean value of 130 HU, respectively. The cystic components of the tumor were not enhanced in these phases, and no pelvic or lumbo-aortic enlarged lymph nodes were observed.

It was confirmed that the mass originated from the left ovarian tissue. Macroscopic examination revealed a

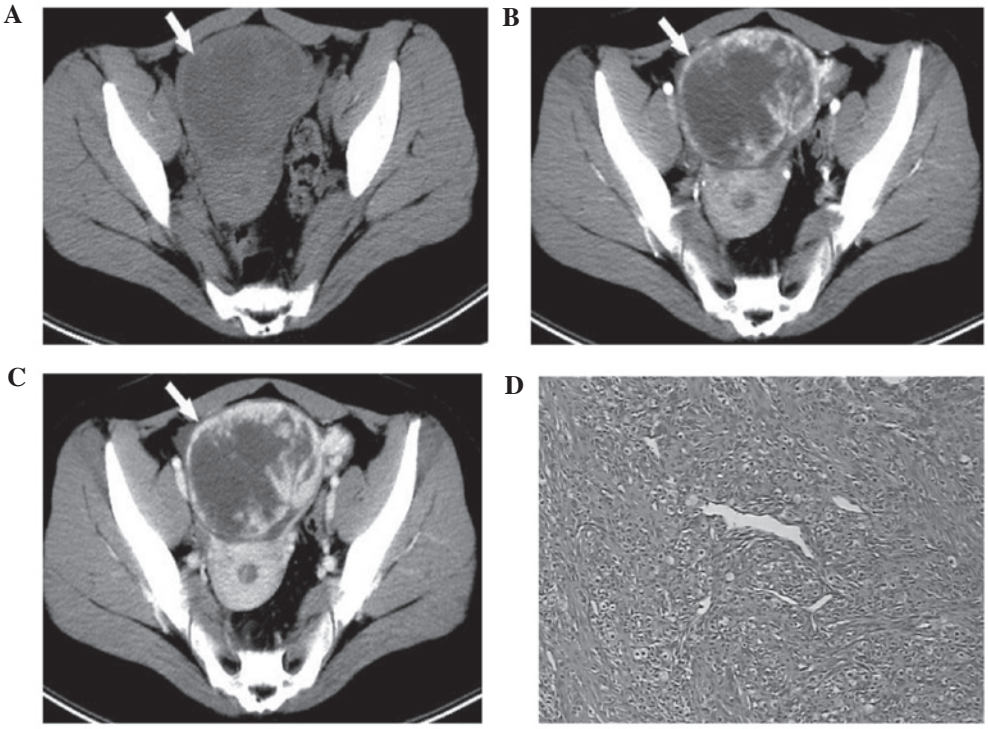

Figure 1. Sclerosing stromal tumor of the left ovary in a 18-year-old woman presenting with menstrual irregularities for 6 months. (A) Axial non-enhanced CT scan showing a round, well-defined heterogeneous mass measuring 76x77 mm in size, which was located in the middle of the pelvis (white arrow). The plain CT values were 31-45 HU. (B) Dynamic contrast-enhanced CT scan showing early peripheral ring enhancement associated with patch enhancement in the center of the lesion (white arrow). On the left side of the tumor, multiple circuitous feeding vessels were clearly visible in the arterial phase. The maximum CT value was 145-150 HU, with a mean value of 147 HU. (C) In the venous phase, the tumor demonstrated progressive, centripetal and prolonged enhancement (white arrow). The CT values were 128-135 HU, with a mean value of 130 HU. The central portion of the tumor was not enhanced. (D) Sclerosing and hypocellular areas separated by edematous fibrous bands were observed with hematoxylin and eosin staining. Blood vessels were mildly ectatic. Magnification, x200. CT, computed tomography.

100x80-mm surgical specimen, which was yellow-orange in color and contained solid, cystic components. Resected tissue specimens were fixed in 10% formalin, paraffin-embedded and cut into 5-mm sections. Sections were stained with hematoxylin and eosin (Shanghai Haiyi Scientific & Trading Co., Ltd., Shanghai, China) for histological examination and staining was visualized using a microscope (JVC1481; Olympus Corporation, Tokyo, Japan). Histological examination of the mass revealed cellular areas separated by hypocellular areas of densely collagenous, edematous or myxoid tissue, alongside prominent vasculature, thus resulting in a pseudo-lobular pattern. The cellular areas were composed of oval to spindle-shaped cells, with a single oval to round-shaped nucleus and a single prominent nucleolus. The cytoplasm was moderately abundant, eosinophilic and occasionally vacuolated (Fig. 1D). Immunohistochemical staining was performed using the avidin biotin peroxidase method, and demonstrated focal positivity for actin, pan-cytokeratin (CK), inhibin and calretinin, and negativity for S100, desmin and epithelial membrane antigen. Based on these findings, a diagnosis of SST of the left ovary was established. The patient exhibited regular menstrual cycles and no signs of local or distal recurrence 2 years post-surgery.

Case 2. A 59-year-old woman was admitted to Subei People's Hospital of Jiangsu Province in May 2013 with continuous pelvic pain that had been ongoing for a year. Clinical examination identified a large mass that was palpable in the right-side of the lower pelvis. Ultrasonography revealed a well-defined solid-cystic echogenic mass in the right ovary measuring 64x55 mm. Laboratory tests and hormonal assays, including CA19-9 (13.11 KU/l; normal range, <35.00 KU/l), carcinoembryonic antigen CA125 (16.15 KU/l; normal range, <35.00 KU/l), α-fetoprotein (1.57 ng/ml; normal range, <20.00 ng/ml), estradiol (31 pg/ml; normal range, 22-144 pg/ml), progesterone (11.13 ng/ml; normal range, 5.16-18.56 ng/ml) and testosterone (21.6 ng/ml; normal range, 10-75 ng/ml) were normal.

Plain CT scan revealed an ovoid soft tissue mass located in the right pelvis. The tumor, which measured 64x55 mm in size, had clear boundaries and showed non-homogeneous density with solid tissue at the periphery (Fig. 2A). Plain CT scan values were 25-40 HU, with a mean value of 32.6 HU. Intravenous bolus injection of iodinated contrast medium yielded early ring enhancement of the peripheral portion of the mass (Fig. 2B). In the venous phase, the degree of enhancement was decreased, but the area of enhancement increased with centripetal progression (Fig. 2C). The CT values at the arterial and venous phases were 125-140 HU, with a mean value of 132 HU, and 118-130 HU, with a mean value of 128 HU, respectively. The cystic components of the inner region of the lesion were not enhanced in these phases.

The patient underwent surgical removal of the 70x60-mm mass, which was well-circumscribed and presented a round to oval-shape with a smooth outer surface. The resected section was of pale yellow color with solid cystic areas. Histological examination revealed that the mass was characterized by

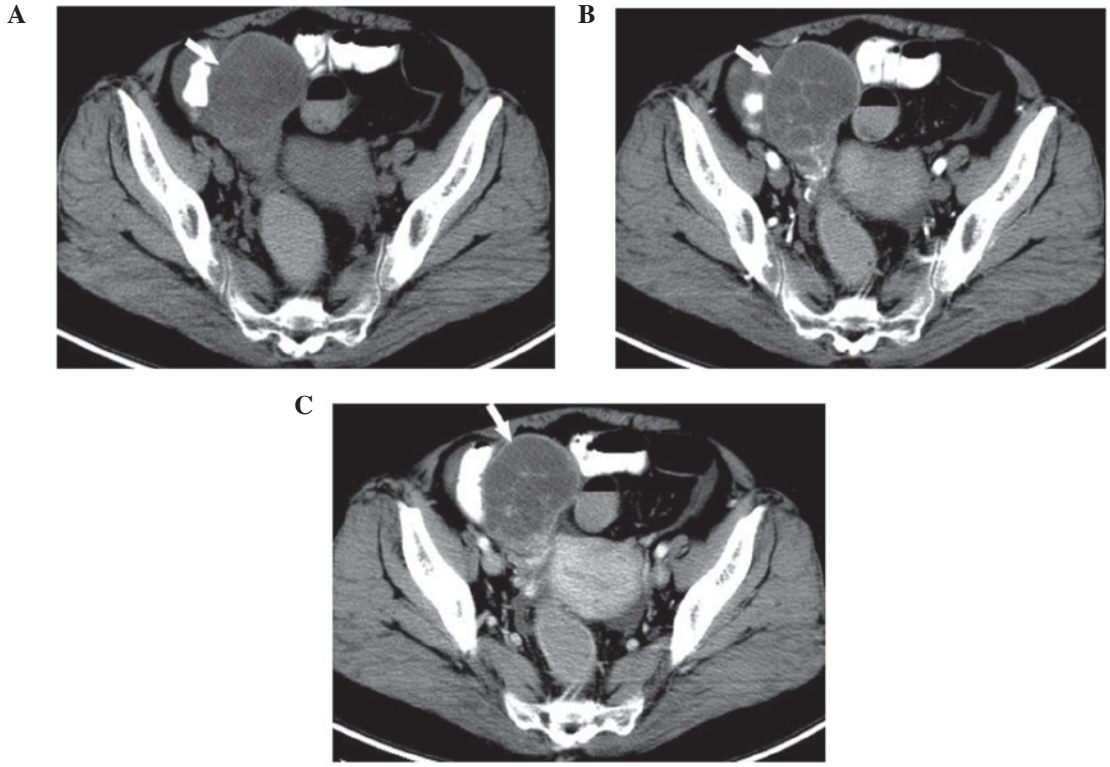

Figure 2. Sclerosing stromal tumor arising from the right ovary in a 59-year-old woman with a history of right-sided pelvic pain. (A) An ovoid solid-cystic mass was observed in the right adnexal area (white arrow). Plain CT values were 25-40 HU, with a mean value of 32.6 HU. (B) Dynamic contrast-enhanced CT scan revealed early septal enhancement in the center, and ring enhancement in the peripheral portion of the mass in the arterial phase (white arrow). The CT value of the solid component was ~132 HU. The CT value of the solid component was 125-140 HU, with a mean value of 132 HU (C) In the venous phase, the tumor demonstrated centripetal, progressive enhancement (white arrow). The CT values were 118-130 HU, with a mean value of 128 HU. The cystic areas of the tumor were observed. CT, computed tomography.

cellular areas separated by hypocellular areas of densely collagenous, edematous or myxoid tissue and prominent vasculature, thus creating a pseudo-lobular pattern. The cellular areas were composed of oval to spindle-shaped cells, with a single oval to round-shaped nucleus and a single prominent nucleolus. Tumor cells stained positive for smooth muscle actin and vimentin, and focally positive for inhibin. The patient was disease-free with no imaging findings of recurrence or metastasis 24 months following surgery.

Discussion

Ovarian SST is a considerably rare and distinctive sex cord-stromal neoplasm, which was first described by Chalvardjian and Scully in 1973 (1). The histogenesis of this tumor remains unknown, and it predominantly affects young women in the second and third decades of life (mean age, 21 years) (2). To the best of our knowledge, <100 cases of ovarian SST have been reported in the English language literature thus far (2-4). Of these, >80% of SSTs have been observed in young adults, in the second and third decades of life (2,3). The most common clinical symptoms of patients with SST of the ovary are menstrual irregularities, pelvic pain and non-specific symptoms associated with the existence of a mass in the pelvic area (2,4). Due to the rarity of this neoplasm, it is not always possible to predict the presence of this tumor preoperatively based solely on clinical and radiological findings (5).

Microscopically, SST is characterized by cellular areas separated by hypocellular areas of densely collagenous, edematous or myxoid tissue and prominent vasculature, which creates a pseudo-lobular pattern (6,7). In a previous study, the cellular areas were observed to be composed of oval to spindle-shaped cells, with a single oval to round-shaped nucleus and a single prominent nucleolus (8). In addition, the cytoplasm was moderately abundant, eosinophilic and occasionally vacuolated (8). A previous study revealed that the blood vessels within the cellular area were mildly ectatic, and no evidence of malignancy was observed (6). Following surgery, the patient's menstrual condition returned to normal (6).

In previous studies, CT imaging demonstrated that the density and enhancement patterns of SST are associated with the cellularity, vascularity, collagen distribution and necrosis or cystic degeneration observed during histopathological examination of the tumor (6,7). On non-enhanced CT, SST manifests solid densities corresponding to cellularity, vascularity and distribution of collagenous or fibrous stroma, while the areas of necrosis or cystic degeneration exhibit low densities (7). The SSTs of the present 2 cases presented patchy areas of low attenuation.

The appearance of SSTs on imaging scans, particularly the enhancement pattern observed in dynamic contrast-enhanced CT images, may vary widely, depending on the distribution of cellularity, vascularity and collagenous or fibrous stroma (9). Previous studies demonstrated that the heterogeneously enhancing masses observed in contrast-enhanced CT images

are associated with the tumor vascularity, cellularity and collagen distribution (7,9). In the present cases, following the administration of intravenous contrast material, the early enhancement of the outer region of the tumor was possibly due to the cellular area with numerous vascular spaces. The maximum enhancement value was 150 HU. In the venous phase, the area of prolonged enhancement observed in the inner portion of the lesion was considered to be associated with the collagenized hypocellular area. The CT values were 110-123 HU, with a mean value of 116 HU. The part of the tumor that did not exhibit any apparent enhancement possibly corresponded to the markedly edematous area. The present CT findings were almost identical to those of previous studies (6,10), and were particularly in agreement with the results of Torricelli *et al* (11), who reported that the appearance of SST on dynamic contrast-enhanced CT images is considerably similar to that of cavernous hemangioma of the liver, and can be pathologically defined as 'hemangioma-like'.

In conclusion, differential diagnosis of SSTs should include other thecoma-fibroma, metastatic and malignant epithelial ovarian tumors (12), since, due to the rarity of SST, prospective imaging diagnosis is not possible.

The present case report described certain imaging features of ovarian SST based on a small-population sample. Therefore, further studies are required to confirm the diagnostic accuracy of imaging in cases of SST, and to determine whether the patterns observed in imaging scans may aid the differentiation between SST and other tumors.

Acknowledgements

The present study was supported by the General Program of Yangzhou Natural Science Foundation (Yangzhou, China; grant no., YZ2015100) and National Natural Science Foundation (Beijing, China; grant no., 81571652).

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