Gynecology and Minimally Invasive Therapy xxx (2016) 1–4

Contents lists available at ScienceDirect

Gynecology and Minimally Invasive Therapy

journal homepage: www.e-gmit.com

Case report

Microcystic stromal tumor resected by laparoscopic surgery

Midori Murakami [a, b, d], Junko Wroblewski [b, c, *, d], Hidehiro Kawagoe [b]

[a] Department of Obstetrics and Gynecology, University of Occupational and Environmental Health School of Medicine, Kitakyushu, Japan
[b] Department of Obstetrics and Gynecology, National Hospital Organization Kokura Medical Center, Kitakyushu, Japan
[c] Department of Obstetrics and Gynecology, Japanese Red Cross Yamaguchi Hospital, Yamaguchi, Japan

ARTICLE INFO

Article history:
Received 15 June 2016
Received in revised form
17 September 2016
Accepted 9 November 2016
Available online xxx

Keywords:
laparoscopic surgery
microcystic stromal tumor
sex cord-stromal tumor

ABSTRACT

We report a case of microcystic stromal tumor (MCST) resected by laparoscopy. MCST is a very rare ovarian tumor with distinctive microcystic features and a characteristic stromal tumor immunophenotype. The present case was a 26-year-old woman who underwent laparoscopic surgery for suspected endometrial cyst of the left ovary. The mass was 8 cm in size and contained bloody fluid, and after attempting cystectomy, we eventually performed left salpingo-oophorectomy with a final postoperative pathological diagnosis of MCST. Although MCST has not yet been associated with malignancy, there are reported links to mutations in the β-catenin gene, and long-term prognosis is still unknown. As MCST resection by laparoscopy has not yet been fully described in the literature, the current case provides an example of when an unexpected, potentially malignant mass is encountered during routine cystectomy and details its subsequent management laparoscopically.

Introduction

Microcystic stromal tumor (MCST) is a very rare ovarian tumor with distinctive microcystic features and a characteristic stromal tumor phenotype, first reported by Irving and Young in 2009.[1] A variant within the sex cord-stromal tumor category, MCST typically affects patients who are 20–60 years old. Tumors are typically unilateral, average approximately 9 cm in size, and have both cystic and solid components.[2,3] Because of its solid component, MCST is often resected by laparotomy, with laparoscopic resection not yet adequately described in the literature.

Here, we report a laparoscopic surgery for a suspected endometrial cyst of the left ovary whose postoperative pathological diagnosis was instead MCST. To the best of our knowledge, this is the first report of MCST resected through laparoscopy, and the current case provides an example of when an unexpected, potentially malignant mass is encountered during routine cystectomy and details its subsequent management laparoscopically.

Case Report

Informed consent for use of patient record and specimens was obtained in written form.

A 26-year-old nulliparous woman presented to our hospital for further evaluation of an abnormal cervical pap smear and dysmenorrhea. There were no complaints of abdominal pain, and personal and family medical history were unremarkable. Upon physical examination, a 6-cm mass was discovered in the left lower abdomen. Transvaginal ultrasound sonography revealed a smooth, thick surface consisting mainly of a cystic component. During internal examination, the tumor was soft, elastic and immovable, with possible adhesions. Magnetic resonance imaging (MRI) revealed a cystic left ovarian tumor with high-signal-intensity appearance on T2-weighted imaging with fluid–fluid level and iso-signal intensity on T1-weighted imaging. There was neither solid component nor contrast enhancement, and MRI revealed a bloody fluid component without any sign of malignancy (Figure 1). The serum tumor markers levels for CA19-9, CA125, and carcinoembryonic antigen (CEA) were within normal limits. Based on chief complaint and imaging results, the diagnosis was endometrial cyst. During outpatient follow-up, however, the tumor seemed to

Conflicts of interest: The authors have no conflicts of interest relevant to this article.

* Corresponding author. Department of Obstetrics and Gynecology, Japanese Red Cross Yamaguchi Hospital, 53-1 Yawatanobaba, Yamaguchi-shi, Yamaguchi-ken 753-0092, Japan.
E-mail address: junkom001@hotmail.com (J. Wroblewski).

[d] Both authors contributed equally to this work.

http://dx.doi.org/10.1016/j.gmit.2016.11.005

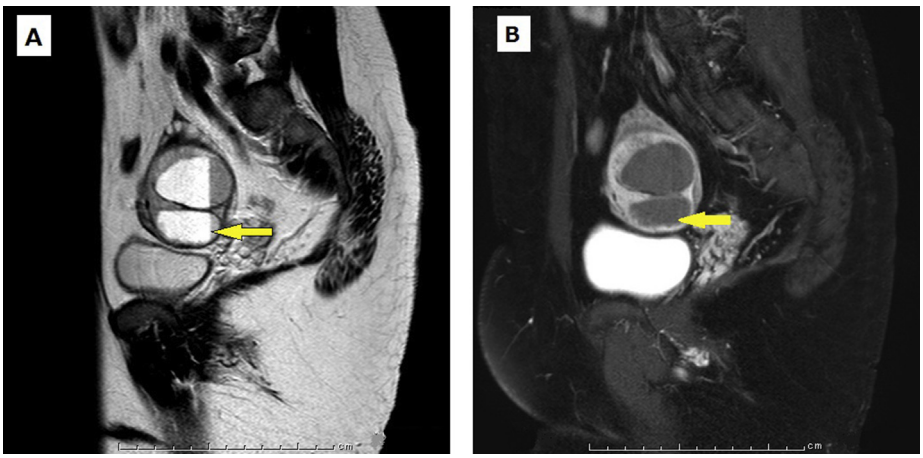

Figure 1. Magnetic resonance imaging. (A) T2-weighted imaging showing a double cystic left ovarian tumor with high-signal-intensity appearance and fluid–fluid level (arrow). (B) T1-weighted imaging showing iso-signal intensity (arrow) in which a bloody fluid component was indicated. There was neither solid component nor contrast enhancement.

have increased in size, and we consequently administered four gonadotrophin-releasing hormone analog (GnRHa) injections at monthly intervals and scheduled a laparoscopic cystectomy.

Four months later, during laparoscopy we discovered an 8-cm cystic mass present on the left ovary without adhesion formation. The uterus and right ovary were unremarkable, with a small amount of serous ascites. For cystectomy, on a part of the mass that was furthest from the oviduct, we chose an area where the tumor wall appeared thick and made a superficial incision in the outermost layer of tissue. From there, we tried to identify the boundary between the normal ovarian surface and tumor layers, but the border was ambiguous and ill-defined, making dissection difficult. In addition, because the tumor surface was also rigid yet fragile, it fragmented easily during the separation process (Figure 2A), rupturing and releasing a bloody fluid. The final resected specimen was thick but brittle, with an irregular surface that suggested the possibility of malignancy. However, definitive determination was difficult midsurgery. Bloody fluid continued to ooze from the abraded area, and neither ligation nor cauterization was effective in stopping blood loss, eventually reaching a total of 500 ml. The procedure was paused momentarily to discuss treatment options with her family that would balance curative treatment and fertility preservation, and after receiving their consent, we finally chose left salpingo-oophorectomy in case any residual tumor remained in the normal ovarian surface layer.

Histopathologically, the tumor was composed of a proliferation of cells with uniform round nuclei and clear or eosinophilic cytoplasm arranged in microcystic, macrocystic, reticulated and solid patterns, and accompanied by myxoid stroma and hemorrhage (Figures 2B–2D). Immunohistochemically, the tumor cells were positive for vimentin (Figure 2F) and CD10, and nuclei were positive for β-catenin (Figure 2E) and FOXL2. Cells were negative for α-inhibin (Figure 2G), as well as for AE1/AE3, CAM5.2, and epithelial membrane antigen (EMA). The MIB-1 labeling index was 8%, and

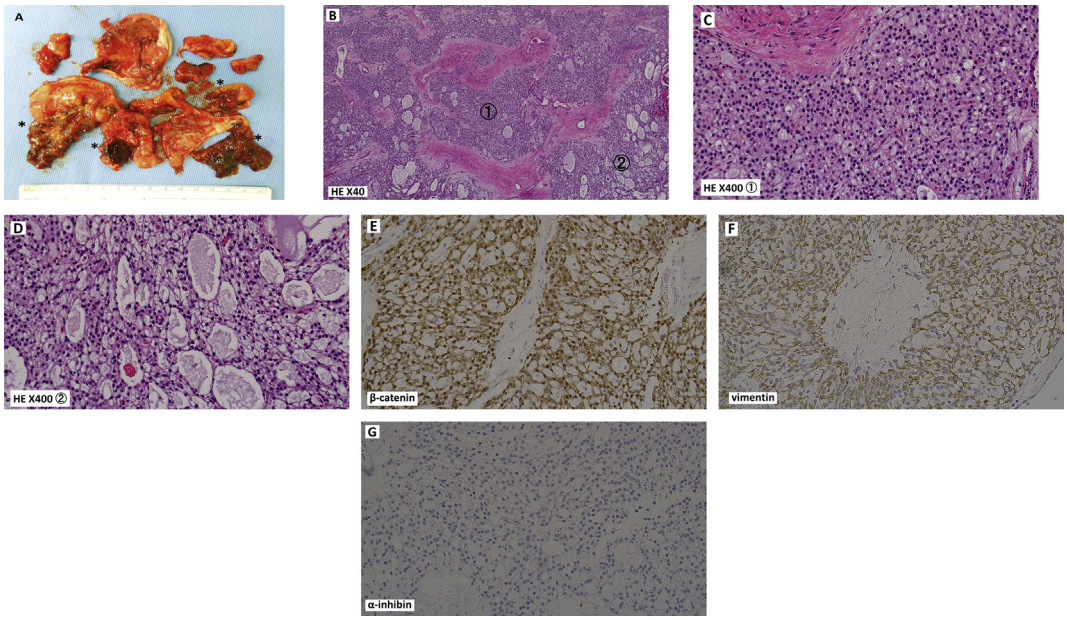

Figure 2. (A) Gross view of the excised specimen. The areas marked by asterisks indicate those areas that were difficult to dissect from the normal ovarian surface. Note that the tumor was fragmented by the dissection process. (B) The different components of the tumor, indicated by ① and ②. (C) Enlargement of ① from B: cells with uniform round nuclei and clear or eosinophilic cytoplasm and a solid pattern with collagenous stroma. (D) Enlargement of ② from B: larger, irregular microcystic, macrocystic, and reticulated component. (E) Immunohistochemistry for β-catenin. Note the positive immunoreactive nuclei. (F) Cells staining positive for vimentin. (G) Lack of immunoreactivity for α-inhibin.

Please cite this article in press as: Murakami M, et al., Microcystic stromal tumor resected by laparoscopic surgery, Gynecology and Minimally Invasive Therapy (2016), http://dx.doi.org/10.1016/j.gmit.2016.11.005