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Sertoli-Leydig cell tumor of the ovary: Radiologic-operative-histologic finding



Given that surgery is the best choice for the management of ovarian tumors, fertility-sparing surgery and use of a minimally invasive technique are important surgical issues in the young adults, and all of these need a comprehensive understanding of the clinical, image, and pathological features of various kinds of ovarian tumors in these young adults to narrow the differential diagnosis and subsequently make an appropriate treatment planning [1]. The following case addressing an uncommon type of ovarian tumor- Sertoli-Leydig cell tumors (SLCTs) of the ovary occurred in a 27-year-old young woman with complaints of menorrhagia for one month.

A 27-year-old woman visited the outpatient clinics due to persistent vaginal bleeding for one month. She had history of thyroid cancer treated with total thyroidectomy with thyroxine treatment at 24 years of age. Transvaginal ultrasound (TVS) revealed a well-defined cystic mass lesion measured with $49 \times 33 \times 56$ mm in size located on the vesicouterine space (Fig. 1). Computed tomography (CT) showed a complex cystic lesion with an enhancing soft tissue part in favor of tumor growth of the left adnexa (Fig. 2 A&B). Physical examination and laboratory data, including tumor markers all revealed unremarkable findings. Then, the patient underwent a laparoscopic surgery for the tumor. During operation, the tumor showed a well-defined mass with yellow-stained and white-colored surface located on the vesicouterine space (Fig. 3A). Since it is nearly impossible to enucleate the tumor and concern the possibility of tumor spillage, laparoscopic salpingo-oophorectomy was performed, and removed through the umbilical port wound with endobag. Macroscopically, the tumor showed multiple dark-red nodules with thick septum and bloody fluid within the ovary (Fig. 4) and microscopically, oval and spindle tumor cells in nested and lobular pattern which were positive for alpha inhibin and calretinin staining but negative for INSM1 (insulinoma-associated protein 1) staining compatible with the diagnosis of SLCT (Fig. 5A–D). Postoperative recovery was uneventful. The patient received postoperative hormone suppression treatment by gonadotropin releasing hormone agonist (GnRH agonist) for maintenance therapy.

To review this case, there are at least three points worthy of our attention. First, the postoperative treatment is still controversial, since SLCTs are rare and typically occur in young women with 75% of patients <30 years of age as well as nearly all patients showed a single ovarian tumor [2,3], suggesting that majority of them did not need further adjuvant therapy. However, based on Taiwanese' experience (a Taiwanese Gynecologic Oncology Group

study), published in 2013, among a total of 23 cases from 9 medical centers in a 20-year period with a diagnosis of SLCTs, 6 patients (26%) needed adjuvant chemotherapy and finally two patients died of diseases [4], hinting us that some of patients will be ended by tumor-related death. Therefore, chemotherapy might not be a better choice for advanced or recurrent SLCTs. Since there are many subtypes of androgen-secreting tumors of the ovary available, such as stromal cell tumors, steroid cell tumors, Sertoli cells, Leydig cells and SLCTs [2,5–7]. In our previous case reports [5–7], we found that the powerful value of the GnRH agonist in the management of the aforementioned androgen-secreting tumors of the ovary. For example, we had successfully used GnRH agonist as a neoadjuvant therapy to suppress the abnormal secretion of testosterone from SLCTs in a 68-year-old high-risky woman, and following laparoscopic surgery for complete tumor excision [5]. We used the same strategy (6-dose GnRH agonist) in the successful treatment for a patient with a biochemical and image-confirmed testosterone-secreting ovarian tumor (renal vein catheterization to collect the serum sample to confirm the secretion of testosterone from the right ovary) due to a poor candidate for surgery in a 70-year-old woman [6]. Final case is a recurrent androgen-secreting tumor in the undetermined sites, which has also been successfully controlled by 4-dose GnRH agonist treatment [7].

Second, although the image could be widely used in the detection of ovarian mass lesions, it is sometimes difficult to use these tools to clarify the rare ovarian tumors, if the tumor was not enlarged. Therefore, many assistance tools might be helpful. One interesting case showing that use of ^{11}C acetate in place of original ^{18}F (fluorine-18)-2 deoxyglucose scanning (FDG) of the whole-body positron emission tomography (WBPET) in the successful detection of recurrent androgen-secreting tumors in 52-year-old woman, who was also successfully treated by a 6-cycle GnRH agonist therapy [8]. There is no doubt that clinical symptoms or signs are a key contributor to hinting us that the patients are possible to have hormone-secreting ovarian tumor [9]. Clinical, ultrasound, CT, magnetic resonance image (MRI), WBPET, and hormone gradient difference of the blood test using either ultrasound-guided, CT-guided, or angiography-blood collecting method or combination of the aforementioned tools can be helpful to diagnose and localize the precise lesion site [8,9]. However, some of tools are not sensitive enough and some are not specific enough. In addition, some of them need the experts we previously commented [10–12]. The hormone gradient difference of the blood test is an example. Therefore, with a high suspicion based on the

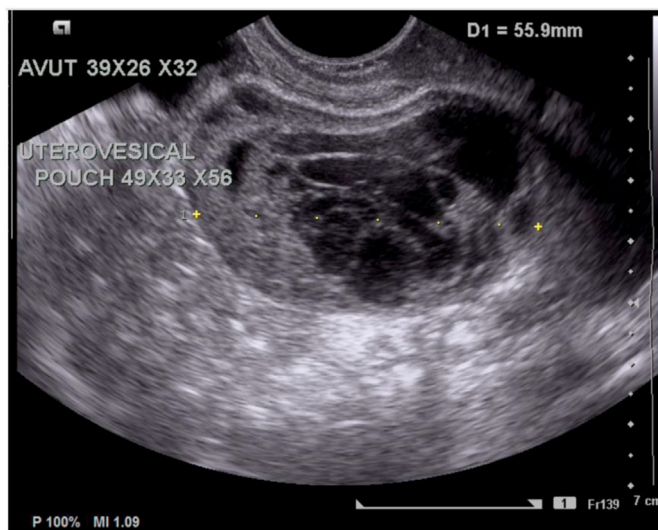


Fig. 1. A well defined complex cystic lesion measured $49 \times 33 \times 56$ mm in size.

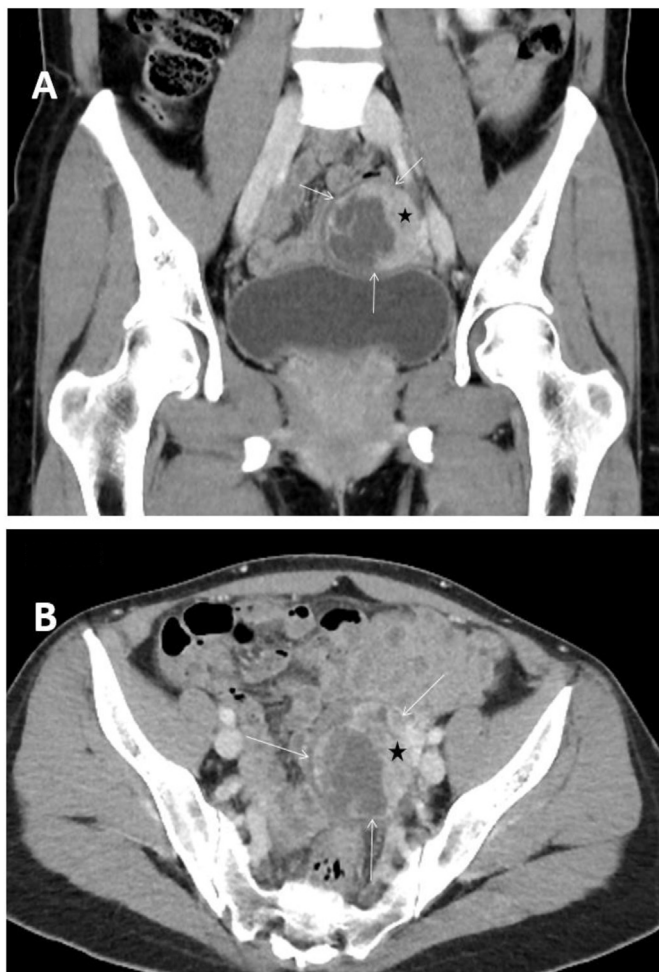


Fig. 2. Contrast enhanced computed tomography: An axial scan in (A) and a coronal reconstruction imaging (B) show a complex cystic lesion (white arrows) with an enhancing soft tissue part in left pelvic cavity, in favor of tumor growth.

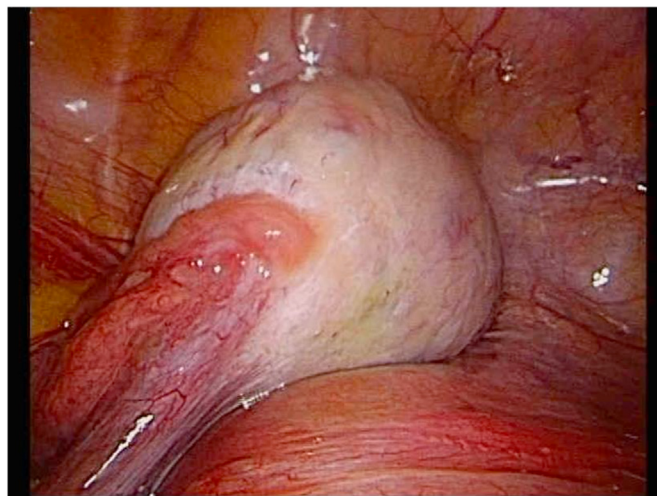


Fig. 3. Laparoscopy showing a tumor located within the vesico-uterine space.

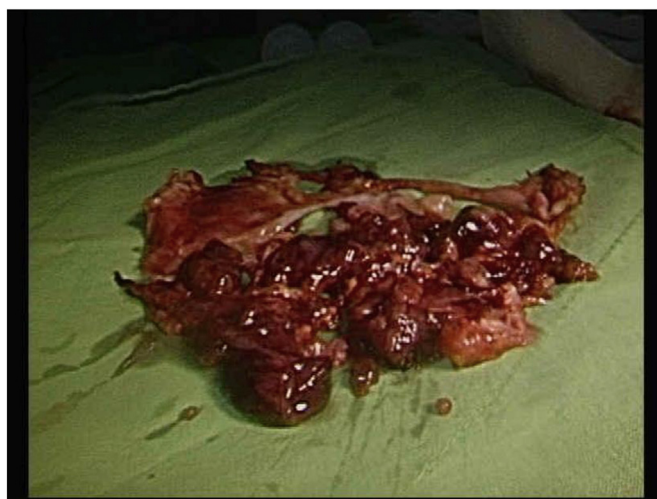


Fig. 4. The tumor revealing lobulated spongy brown sectioned surfaces.

availability of clinical data, therapeutic trial with a relatively safe treatment, such as our previously mentioned GnRH agonist might be an alternative [5–7].

Third, although patients with testosterone-secreting ovarian tumor, in theory, will present with androgen-related clinical symptoms or signs, many will present symptoms or signs not related to excessive androgen effects. In literature review, only half of patients present with hyperandrogenism [2,3]. However, since this-type tumor shows various degrees of hormonal abnormalities, including irregular menstruation, abnormal vaginal bleeding, etc., just like our current presented case, suggesting that women or girls complaining of menstrual problems, the possibility of functional tumors should be taken into consideration. Since majority of them can be successfully managed by complete surgical excision, the omission should be avoided.

Taken together, the current case is interesting and highly educative. The shared experience will enhance the better care of patients [13].

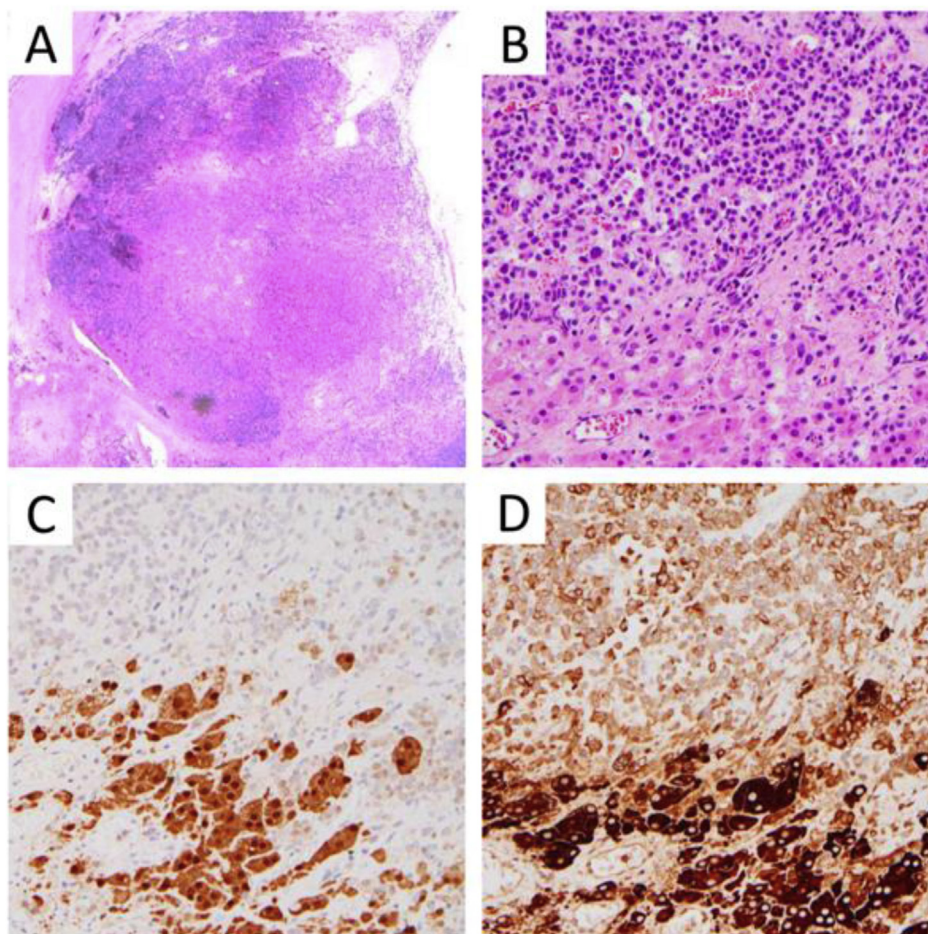


Fig. 5. Ovarian Sertoli-Leydig cell tumor, intermediate differentiated. (A) The tumor is arranged in nodular architecture with cystic change and edematous stroma; H&E, x20. (B) Tubules and cords of Sertoli cells are noted in the upper part, whereas nests of Leydig cells with abundant cytoplasm are seen in the lower part; H&E, x200. (C) Calretinin immunostaining is positive and relatively stronger in the Leydig cell compartment; x200 (D) Alpha-inhibin immunostaining is diffusely positive and shows stronger expression in the Leydig cells than the Sertoli cells; x200.

Declaration of competing interest

The authors declare that they have no conflicts of interest related to the subject matter or materials discussed in this article.

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