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Case Report

Borderline Ovarian Tumor in Children and Adolescents, a Case Report: A Rare Tumor

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Abstract

Borderline serous ovarian tumors represent less than 1% of ovarian tumors in adolescents. The diagnostic approach includes clinical examination, imaging, and tumor markers. The definitive diagnosis is anatomopathological. Treatment is surgical while preserving fertility. Borderline tumors have a good prognosis with a risk of recurrence. Our work concerns the management of a case of a borderline ovarian tumor in a 16-year-old adolescent.

Keywords: *Borderline Ovarian Tumor; Children; Adolescents*

Introduction

The diagnosis of ovarian tumors in pediatrics and adolescents is often difficult and delayed due to non-specific warning signs and polymorphic presentations in imaging (pelvic mass). These tumors are rare, estimated in children and adolescents at approximately 2.6/100,000 girls per year, functional lesions being excluded [1,2]. 10 to 20% of them are malignant [1].

However, ovarian tumors remain poorly understood in this age group. To this end, we report a case of ovarian tumor in a 16-year-old adolescent girl, in order to analyze the diagnostic value of the signs observed and to propose a diagnostic orientation in the face of the discovery of an ovarian mass syndrome in children or adolescents.

Patient and Method

Clinical semiology

The symptomatology related to a pelvic mass in children and adolescents is polymorphic [1].

This was a 16-year-old patient who consulted for a pelvic mass. The patient was from a consanguineous marriage, menarche at 9 years old with regular cycles. The symptomatology was limited to chronic pelvic pain of the heaviness type, the evolution was marked by the appearance of a pelvic mass that increased in volume. On clinical examination, we noted the presence of a hypogastric mass reaching 2 finger widths below the umbilical, firm and painless to palpation without other associated physical signs.

Radiological explorations

The radiological exploration included: Standard radiography (Abdomen without preparation), ultrasound, computed tomography (CT), magnetic resonance imaging (MRI) [1].

Abdomino-pelvic ultrasound

In the literature, abdomino-pelvic ultrasound is the first-line examination [2,3]. It allowed to locate the pelvic mass, study its characteristics, specify the organ of origin and analyze its aggressiveness [1].

Abdomino-pelvic ultrasound was the first complementary examination performed on our patient. It objectified a roughly oval, well-defined suprauterine hypogastric mass, with bumpy contours measuring 120 × 80 × 130 mm. This mass is isoechoic with an anechoic center, vascularized on Doppler. It is directly related to the anterior parietal peritoneum, it pushes the uterus down, the uterus is of normal size, thin endometrium, homogeneous myometrium, the right ovary was visible without abnormality, the left ovary is hidden by the mass. The bladder is pushed downward with loss of the separating fatty interface (Figure 1 and 2).

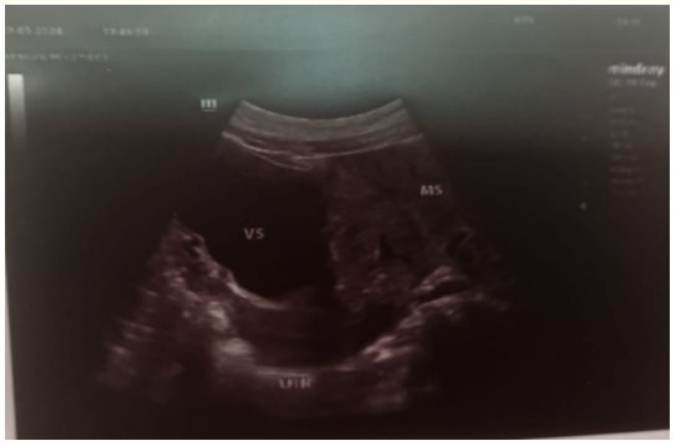

Figure 1: *Pelvic mass coming into contact with the bladder without loss of separation line.*

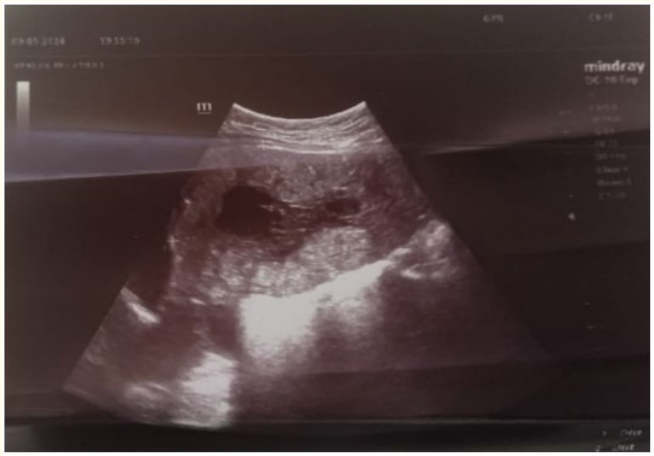

Figure 2: *Roughly oval, well-defined suprauterine hypogastric mass with bumpy contours. This mass is isoechoic with an anechoic center.*

Abdomino-pelvic MRI

In front, the impossibility of specifying the origin of the pelvic mass by ultrasound. CT and especially pelvic MRI could provide useful information [1,3].

It objectified a tissue abdomino-pelvic mass, located above the bladder, slightly lateralized to the left, oval, measuring 73 × 145 × 116 mm, well-defined, with regular contours, in T1 hyposignal, in T2 heterogeneous hypersignal, in diffusion hypersignal, with restriction of the apparent diffusion coefficient (ADC), significantly enhanced by the contrast product (PDC) and site of a central liquid zone. At the bottom, it comes into intimate contact with the bladder dome and the uterine fundus with loss of the separating fatty interface. Behind, it comes into intimate contact with both ovaries with loss of the fatty interface of separation and without any obvious sign of ovarian affiliation. Anteriorly, it comes into intimate contact with the muscles of the left anterior and lateral abdominal wall. Above, it comes into intimate contact with the small intestine loops with loss of the fatty interface of separation in places. Behind, it comes into intimate contact with the left psoas muscle, the aortic bifurcation and the right and left primitive iliac vessels and the left internal and external iliac vessels which remain permeable, the sigmoid and in contact with L5 with persistence of a thin fatty border of separation. The ovaries are of normal follicular size, the uterus of normal size with regular contours with respect for its zonal anatomy, the bladder of good capacity with thin wall, with respect for the parietal hyposignal, absence of thickening of the rectal wall, absence of suspicious adenopathy. Low abundance pelvic effusion (Figure 3 and 4).

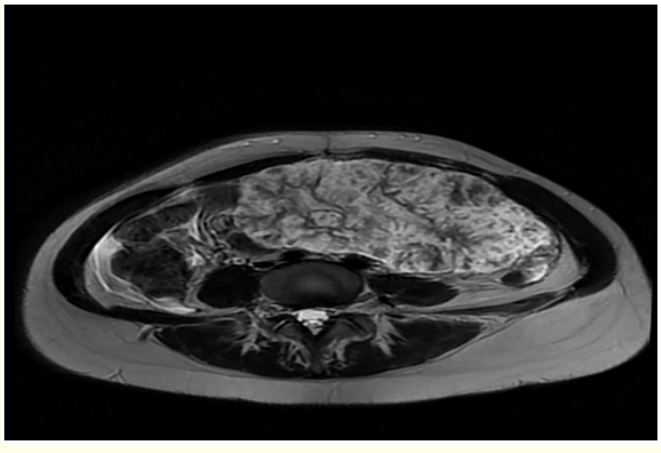

Figure 3: *Abdominopelvic mass, located above the bladder, slightly lateralized to the left, oval, measuring 73 × 145 × 116 mm, well defined, with regular contours.*

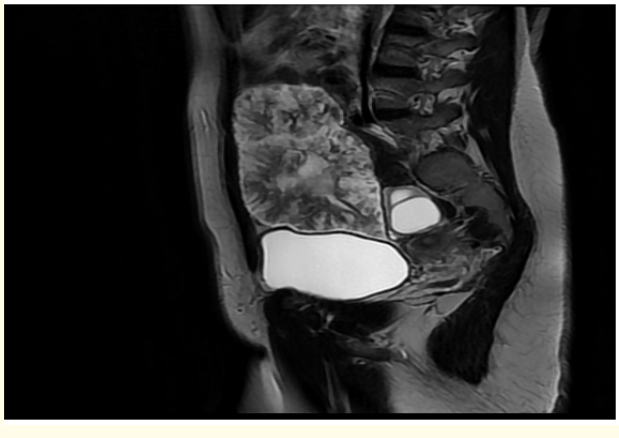

Figure 4: *Abdominopelvic tissue mass, located above the bladder, slightly lateralized to the left, oval.*

MRI allowed visualization of intratumoral blood as hypersignal in T1-weighted sequences that disappears after fat suppression, this pointed towards a hemorrhagic cyst, a hemorrhagic ovarian infarction, juvenile granulosa cell tumor. Our examination found a central fluid area not corresponding to the bleeding. Uterine abnormalities, particularly endometrial, pointed towards granulosa cell tumors, no uterine abnormality was noted in our radiological examinations [1].

Another interest of MRI is the search for metastasis [1]. In our case, the tumor appeared locally aggressive, without adenopathies.

Biological explorations

Tumor markers are the essential biological element for the management of ovarian masses suspected of malignancy [1,3]. Among these markers, alpha-fetoprotein (AFP) and the ß branch of human chorionic gonadotropin (ßhCG) had a sensitivity of 78%, a specificity of 100%, a positive predictive value of 100% [1,2]. Inhibin is a useful tumor marker in germ cell tumors, stromal tumors and sex cord [1,3,4].

In our case, we were satisfied with the dosage of ßhCG which was negative.

CA 125 is another interesting marker in ovarian masses; In our case, the initial value of CA 125 exceeded 600 IU/ml.

Surgical exploration

In our case, imaging objectified the pelvic mass and its aggressive nature without specifying its origin, with the high CA125 level, surgical exploration was of great interest.

This exploration had a diagnostic, prognostic and therapeutic purpose.

The patient was scheduled for an exploratory laparotomy. Intraoperatively (Figure 5 and 6), we noted the presence of a vesicular vegetating pelvic mass at the expense of the left ovary measuring 16×14 cm. We also noted the presence of multiple nodules of peritoneal carcinomatosis, the omentum and the contralateral ovary. The left fallopian tube was adherent to the rectum and to the pelvic wall with the presence of adhesion from the rectum to the posterior wall of the uterus and the vagina.

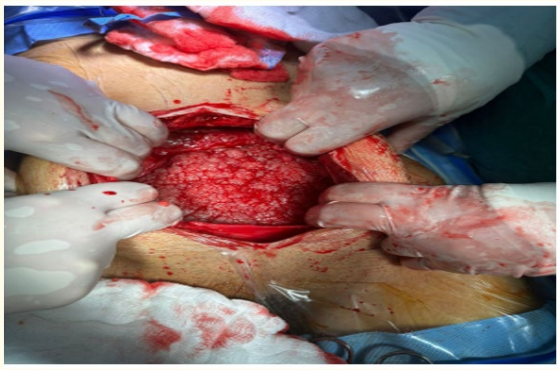

Figure 5: *Abdominopelvic vesicular mass.*

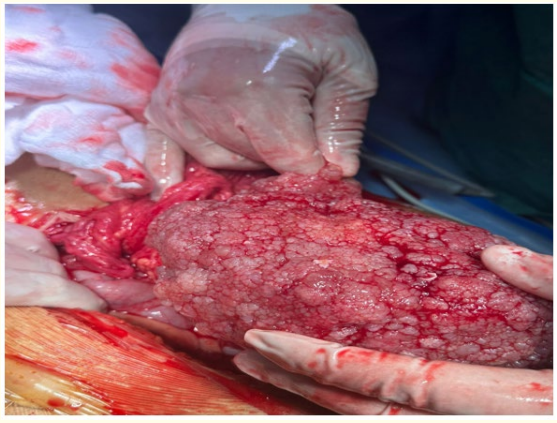

Figure 6: *Vesicular vegetating pelvic mass at the expense of the left ovary measuring 16×14 cm.*

The patient underwent a left ovariectomy associated with multiple biopsies (contralateral ovary, omental, peritoneal) and peritoneal cytology.

Anatomopathological result

The WHO classifies ovarian tumors into three main groups depending on whether they are derived from the surface epithelium, germline cells or ovarian stroma or sex cords [1]. Primary or secondary localizations are possible in malignant hematological diseases [1].

On anatomopathological study: Left ovariectomy specimen, site of a borderline serous tumor, remodeled, without invasive character. It is associated with a non-invasive omental implant. The contralateral ovary and peritoneal tissue are free without tumor localization. Peritoneal cytology was negative.

Discussion

The annual rate of ovarian neoplasms in children and adolescents is estimated at 2.6 cases per 100,000 girls, which illustrates the rarity of this condition [1]. They are generally benign and the most common types are mucinous cystadenomas, mature teratomas and serous cystadenomas (Table 1) [1].

Lesions	Benign	Borderline	Malignant
Germ cell tumors	Mature teratomas		Seminoma Yolk sac Immature teratomas Mixed teratomas
Sex cord and stromal tumors	Sclerosing stromal tumors		Juvenile granulosa cell tumors Sex cord tumors with annular tubules
Epithelial tumors	Mucinous cystadenomas	Mucinous cystadenofibromas Mucinous intestinal_type tumors	
Other tumors			Mixed unclassifiable sex cord and stromal tumors
Non-tumoral lesions	Hemorrhagic cyst Pseudotumoral hemorrhagic infarction		

Table 1: *Distribution of different ovarian tumors [1].*

The symptomatology related to an ovarian mass is polymorphic (Table 2), it can be asymptomatic or even go as far as an increase in abdominal circumference [1]. It can also be accompanied by signs and symptoms suggesting compression, such as nausea, vomiting, weight loss or increased urinary frequency, urinary retention, constipation and dyspnea [1]. Acute pelvic pain required suspicion of adnexal torsion and then emergency laparotomy [1,2]. In our case, it was a chronic symptomatology. Pelvic pain and the palpable mass observed in our patient are the major symptoms reported in the literature (Table 2).

Symptoms	%
Abdominal pain	78
Palpable mass	56
Distended abdomen	39
Nausea/Vomiting	36,5
Fever	12
Asthenia	10
Early puberty	7
Post-puberty metrorrhagia	2,5
Virilisation	2,5
Hemorrhagic shock	2,5
Chance discovery	2,5

Table 2: *Revealing symptoms [1].*

It is of paramount importance for ovarian masses to establish the probability of malignancy prior to surgical intervention [1,2]. In order to assess this risk of malignancy, all patients should undergo a complete physical examination as well as imaging studies and evaluation of serum tumor markers [1,2].

Ultrasound is the primary imaging method used in the evaluation of an ovarian mass [1,2]. It can provide information on the size and volume of the cyst, as well as its morphology (the wall of the cyst, whether it is unilocular or multilocular, or whether it contains solid elements) [1,3]. Doppler examination is useful to evaluate the vascular component in the solid parts of the tumor or in the septa [1,2].

In clinical practice, some ultrasound features can guide the diagnosis: For example, cystadenomas are unilocular cystic masses without solid components, in mucinous cysts, different fluid densities can be observed due to mucin stratification. Mature teratomas have both liquid and solid components with calcifications, echogenic shadows of sebaceous material and levels of adipose fluid. Posterior shadows of solid parts are characteristic of these tumors. In contrast, malignant tumors tend to present as masses with ill-defined and irregular contours, with central necrosis, thick and vascularized septations and papillary projections. They present ovarian vegetations or intracystic vegetations [1].

Ultrasound indicators of malignancy include solid components larger than 2 cm, thick and irregular septations, papillary projections with Doppler signal and ascites [1]. In our case, ultrasound demonstrated a pelvic mass larger than 2 cm, isoechoic with an anechoic center, vascularized on Doppler, exerting a mass effect on the bladder and uterus.

However, it is not always easy to establish the ovarian origin of tumors. MRI and CT can provide useful information [1]. In our case, MRI provided a more precise study of the ovarian mass and its extension without specifying its origin.

Regarding relevant tumor markers, the most commonly evaluated are CA125, AFP, ßhCG, LDH, and inhibin A and B [1,2]. Tumor markers may not be elevated in early-stage ovarian neoplasia, but they can be useful in the evaluation of ovarian tumors with a high rate of suspicion of malignancy [1]. By taking into account the levels of the markers, preoperative planning of a staging procedure can be easy and postoperative follow-up can be performed [1,2].

The high level of CA125 in our case raised suspicion of malignancy of the mass.

At the surgical stage, the therapeutic objectives are: remove the mass without leaving any residue, improve symptoms, perform staging, preserve fertility and avoid complications [2].

Laparotomy is superior to laparoscopy given the low risk of rupture during surgery, the low risk of recurrence, the ease of palpation of areas suspected of malignancy [2,3]. The surgical procedure is cystectomy or oophorectomy [1]. Ovariectomy is the treatment of choice if the ovarian lesion is > 8 cm, or there is a risk of malignancy [3,4]. Zhang., *et al*. also described a significant difference in tumor size between patients with benign neoplasia (5.7-9 cm) and those with malignant tumors (8.6-17,3 cm) [5]. Peritoneal biopsy and cytology, biopsy of suspicious lesion [2,3,5].

In our case, we opted for a median laparotomy infra-umbilical enlarged supra-umbilical for a better exploration allowing to specify the ovarian origin of the pelvic mass. A left ovariectomy was performed given the dimensions of the mass observed on imaging, a sample of peritoneal fluid for cytological study with biopsy of suspicious peritoneal nodule, suspicious omental nodule and suspicious contralateral ovarian nodule.

The anatomopathological result: The left ovariectomy specimen contains a borderline serous tumor without invasive character. The peritoneal biopsy, the omental biopsy and the biopsy of the right ovary are free of tumor localization. The peritoneal cytology is not inflammatory and without suspicious cells.

The anatomopathological result: The left ovariectomy specimen is the site of a borderline serous tumor without invasive character. No tumoral localization on the peritoneal biopsy, the omental biopsy and the biopsy of the right ovary the peritoneal cytology is not inflammatory and without suspicious cells. There is no consensus on postoperative follow-up, chemotherapy is avoided as much as possible in order to preserve the patient's fertility [3]. However, borderline ovarian tumors have a risk of recurrence of 43% [2]. Some propose post-operative monitoring by pelvic ultrasound and plasma dosage of CA 125 every 3 months during the first year then every 6 months during the second year then annual monitoring [6], others opt for clinical and paraclinical monitoring (tumor marker dosage and pelvic ultrasound) every 3-6 months during the first 5 years then annual monitoring for 10 to 20 years [3]. We decided to follow our patient post-operatively in consultations every 3 months during the first year. This monitoring included: clinical signs, CA 125 kinetics by monthly dosage for the first 3 months then every 3 months if there was a good decrease, ultrasound examination. Our patient is currently asymptomatic without ultrasound signs of recurrence with a decrease in CA 125 levels.

Monitoring rate	Pre-operative	1 month postoperative	2 month postoperative
CA125 rate	>600 UI/ml	54 UI/ml	8 UI/ml

Table 3: *Kinetics of CA125.*

Algorithm of medical care

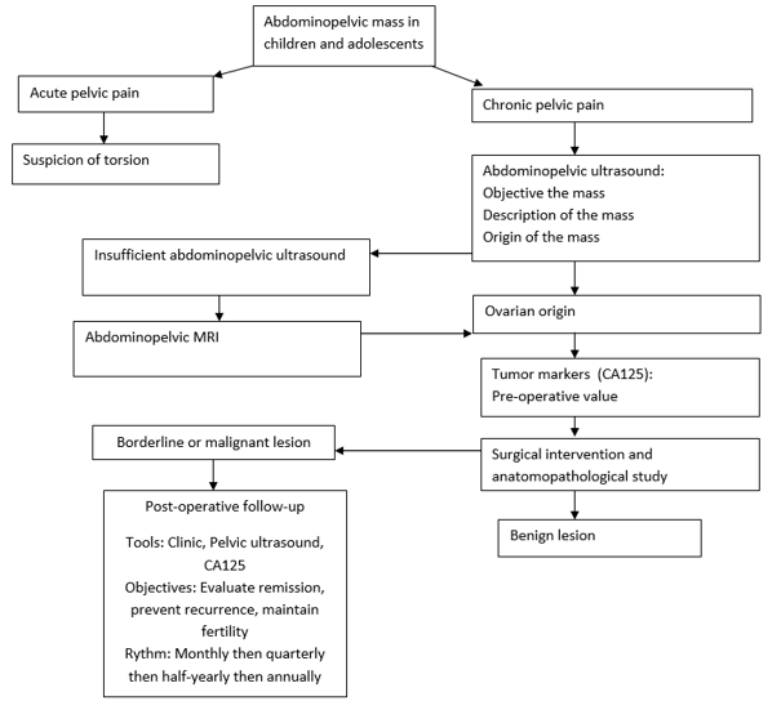

Figure 6: *Vesicular vegetating pelvic mass at the expense of the left ovary measuring 16×14 cm.*

Conclusion

Our work aims to report a case of a borderline ovarian tumor in an adolescent, with the ambition of proposing a diagnostic, therapeutic and monitoring strategy. The main symptom is the abdominopelvic mass, imaging is essential for the etiological diagnosis, ultrasound first. Surgery remains the adequate treatment. Tumor markers and imaging are useful in case of suspicious ovarian mass. Post-surgical follow-up aims to assess remission, detect recurrence and preserve fertility.

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