RARE OVARIAN TUMOUR ASSOCIATED WITH SECONDARY AMENORRHEA. CASE REPORT

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Abstract: *This paper aims at presenting the steps to diagnose and to laparoscopically treat some rare ovarian pathology – voluminous ovarian fibromyoma complicated with secondary amenorrhea, in a teenager.*

INTRODUCTION

The ovarian fibroma is a solid benign tumour, the most frequent of the ovarian tumours that originates in the non-hormonal cells of the cortical ovarian stroma. Sometimes, some of these cells can have hormonal activity, in this case the tumours being named fibrothecomas. These tumours are rare in the premenstrual period, though Chen and co. described such tumours in girls as young as 7 years old.(1) In 1% of the cases, the ovarian fibroma can be associated with ascites and pleural effusion, mostly on the right side and altogether they are forming the Demons-Meigs syndrome.(2,3)

CASE REPORT

We present the case of a young, healthy, 17-year-old patient, student, who comes to the clinic with pelviabdominal pain, discomfort and tenesmus.

From the physiological case history it is important to keep in mind the late menarche, at 17 years, with 3 irregular menses in 9 months, with variable duration (3-10 days) with marked dismenorrhea. The patient does not have a sexual history.

The current disease had an abrupt setting three month ago with pelviabdominal pain and tenesmus, and became worse in the last days.

The clinical exam reveals normal aspect breasts, corresponding with age, flat, supple abdomen, painful in the hypogastric region with posterior radiation, with preserved intestinal transit and digestive tolerance.

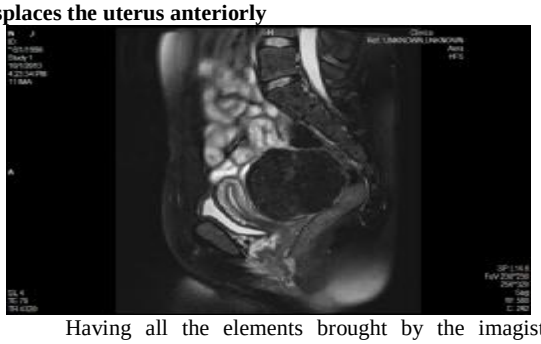
Being virgo intacta, we can only perform the pelvic examination by rectal exam. We can palpate the uterus in an abnormal position (hyper anteverted anteflexed uterus) because of a firm tumour, well delimited with slightly irregular margins, mobile on the neighbouring structures, supple adnexal lodges, the Douglas pouch being occupied by the tumour. The rectal ampula is partially compressed by the tumour.

To be able to support a positive, etiological and differential diagnosis we have performed paraclinical, usual and special laboratory investigations. The ordinary investigations were found to be in normal limits, as well as the weight, blood pressure, pulse, temperature curves and diuresis.

We have performed a genital ultrasound that reveals the uterus, 68/15 mm, with homogeneous echostructure, moved anteriorly by a hypoechoic tumour, well delimited, 6 cm in diameter. The aspect of the left ovary is normal. We cannot highlight the right ovary.

The MRI examination describes a bulky mass in the pelvis (77/56 mm), slightly polycystic, well delimited, hyperintense in T2. These characteristics are consistent with fibromyoma. The tumour is situated on the posterior wall of the uterus which is moved over the bladder, without modifying its structure. The situation between the uterine corpus and the rectal ampula suggests a pediculated, subserosal uterine origin. The aspect of both ovaries is normal, with well highlighted de Graaf follicles.

Figure no. 1. Retrouterine tumour, well delimited, that displaces the uterus anteriorly



Having all the elements brought by the imagistic investigations we have diagnosed the patient with pediculated, subserosal uterin fibromyoma complicated with compressive phenomena.

We decided to approach this tumour through laparoscopic surgery in order to confirm the diagnosis and to perform the myomectomy.

Figure no. 2. The tumour that occupies all the pelvic region, 10/6 cm, very well delimited, hyperintense in T2

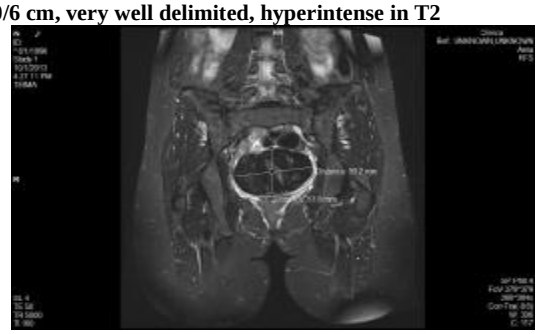


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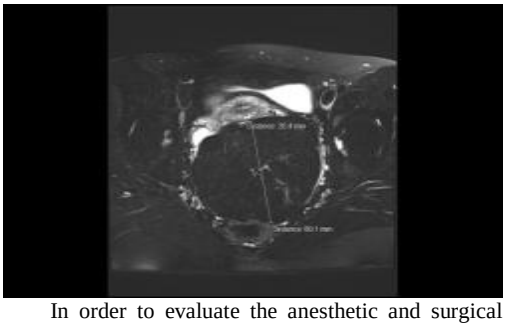
Figure no. 3. Comparison between anterior-posterior diameter of the tumour and the same diameter of the cervix



In order to evaluate the anesthetic and surgical risk factors we have ordered a clinical and biological balance sheet, cardiopulmonary X-Ray and ECG, along with preoperative preparation: preanesthetic fasting, enema and bladder catheterization.

For this kind of surgery there was need for general anesthesia with orotracheal intubation.

During the surgery we have found the normal appearance uterus and the left adnexa, the right ovary with a tumour-like appearance, 6-7 cm, pearl white, firm, that looks like a solid ovarian tumour, situated posterior to the uterus, with no free fluid in the peritoneal cavity, no signs of pelvic endometriosis and normal aspect for all the intraperitoneal organs.

We decided to perform right laparoscopic adnexectomy. We have clipped, coagulated and sectioned the right infundibulopelvic ligament, the proper ovarian ligament and the Fallopian tube near the uterus. Because of the firm, bony consistency of the tumour enforced us to expand laterally the hypogastric trocar incision for 4 cm. We have performed haemostasis control, peritoneal washing, exsufflation of the pneumoperitoneum and the suture of the trocar incisions.

Figure no. 4. Excised right ovary

The specimen is sent to pathology for the histopathological exam.

There were no incidents and accidents in the immediate postsurgical and postanesthetic period.

After the surgery we ensured good analgesia, infection prevention through antibioprofilaxy, surgical wound toilet and early removal of the urinary catheter, deep venous thrombosis prevention through pharmacological and mechanical measures (early mobilization of the patient) and fluid management.

The patient was discharged from the hospital the third day after the surgery in good general condition, afebrile, resumed intestinal transit and good digestive tolerance, spontaneous micturition, the surgical wounds in process of healing.

The menses resumed regularly after the surgery.

Pathology result: ovarian leiomyoma with calcification areas.

DISCUSSIONS

We have reported a case of ovarian fibroma that raised problems of clinical and imagistic differential diagnosis because of the exclusion of genital examination and endovaginal ultrasound, the patient being virgo intacta.

The etiopathogenesis of the ovarian leiomyomas is unknown, though isolated studies suggest that a genetic component can be described. There were found high levels of mRNA aromatase in the leiomyomas or a high prevalence of genetic polymorphism in the estrogen receptor gene in the Afro-American women.

The differential diagnosis of the histological type for the solid ovarian tumours is made with:

- Uterine benign tumours: other types of uterine fibromyoma are infirmed by the IRM examination that describes the endometrial cavity, endometrium and myometrium with normal appearance.
- Solid ovarian tumours like Brenner tumour (the epithelial cell isles are missing in the fybroma), dysgerminoma or other type of tumours that present with areas of calcification, like dermoid cysts, serous cystadenomas, cystadenocarcinomas, gonadoblastomas, lesions after tissue necrosis.

The particularity of the case consists in the presence of amenorhea associated to an ovarian fibromyoma in a teenage patient, without any risk factors for this kind of tumour (early menarche, obesity, Afro-American, polycystic ovary syndrome). The only risk factor that could stand is the genetic one, but it is not documented.

The fibromyomas represent 5% of all ovarian tumours, being most frequently encountered in the menopausal period. Though it is considered a non-secreting tumour, it can be accompanied by sterility (6), menstrual disorders, compressive disorders, Meigs syndrome.

When the ovarian fibromas are found bilaterally, calcificated, multinodular and associated with basocellular skin cancer, they compose the basocellular nevus syndrome. The ovarian fibroma can also be associated with ascitis and hirsutism.(7)

Macroscopic, 90% of the fibromas are unilateral and pediculated, variable in size, smooth surface, with the tendency to conserve the shape of the ovary. On section, they are solid, firm, with areas of calcification given the repeated episodes of ischemia following repeated twists around the pedicle (10%).

The ovarian fibroma can display hormonal receptors for estrogen and progesterone. There are clinical syndromes that are characteristic for the receptor presenting tumours: ovarian fibroma that is associated with ascitis and hydrothorax compose Demons – Meigs syndrome.

The association of the fibroma only with ascitis is more frequent. In 50% of the cases, the ascitis is an intraoperative discovery. The pathogenesis of Meigs syndrome is though to be the inadequacy of the lymph drainage through a pedicle that is to narrow for the growing tumour mass. In addition, the tumour and the ovary as well do not have a proper capsule to prevent the lymph leakage in the peritoneal cavity. The pleural liquid results from ascitis migration through transdyaphragmatic lymphatics. The complete Meigs syndrome can be encountered in 1% of the ovarian fibromas; 40% of fibromas over 6 cm are associated with ascitis. The biggest statistic belongs to Gargano, with a number of 34 cases reported in 2003.

The evolution of the leyomyomas depends on their

size, location, the development direction, age, physiological state (pregnancy, childbed). Generally, the disease has a slow progress; in this case the tumour got bigger, increasing the risk to twist – important cause of acute surgical abdomen or slow twist – cause of aseptic necrobiosis.

The complications can be local, mechanical, through compression on the neighbouring organs or through acute or slow twisting; local vascular complications edematous degeneration or aseptic necrobiosis; local benign degenerative complications or malignant sarcomyoleiomatosis.

The complications can also be classified as surgical, intra- or postsurgical. During the surgery we mention the injury of the neighbouring organs, haemorrhage, and after the surgery we can expect hematomas, mechanical obstructions through bridles, infections.

The ultrasound examination describes a solid tumour, well defined, hypoechoic, rarely hyperechoic or anechoic. Through tomodensitometry and MRI, the tumours can be differentiated in benign and malignant.(5,6) The endovaginal ultrasound is very useful for very small tumours. The colour Doppler ultrasound is useful for increasing the accuracy of the ultrasound to differentiate the benign from malignant tumours.

Another investigation used to differentiate the tumours is the dosing of CA-125, especially for women at reproductive age. A value of this marker higher than 200U/mL in the presence of an adnexial tumour (especially after the menopause) is most probably associated with malignancy.(7)

The best preoperative diagnostic method, especially for the small tumours, seems to be the association of the endovaginal ultrasound with dosing the CA-125 marker.

Some authors believe that the aromatase inhibitors can reduce the dimensions of the uterine or ovarian fibromas (10), but the definitive treatment remains surgery, classic or laparoscopic. At reproductive age, the surgical attitude should be conservative with the maintenance of the reproductive function. After menopause, total hysterectomy with bilateral adnexectomy is recommended in some cases. In 1% of the cases the tumours that are supposed to be benign are proved to be malignant ovary tumours.(7)

The extraction of the big solid tumours, like the ovary fibrothecoma, in the laparoscopic technique, is done with the help of an endobag through an enlarged trocar incision; rarely, the little tumours can be extracted at the same time with the histerectomy piece vaginally or through posterior colpotomy.(7)

The vital prognosis is good if the surgical treatment is applyed correctly and on time, but when the complications start to appear, especially the malignancy, the prognosis becomes worse. The functional and reproductive prognosis is not affected if there was a conservatory treatment performed. The recurrence of the disease is very rare.

CONCLUSIONS

The extremely small number of cases in this age category and the lack of reports in the recent literature reflect the rarity of this pathology. In the reported case, another particularity is represented by the fact that the ovarian fibroma was associated with secondary amenorrhea given that the other ovary had a normal appearance.

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