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Case Report

A case of ovarian juvenile granulosa cells tumor with peritoneal and jejunal localisation at Amath Dansokho Regional Hospital, Kedougou, Senegal

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ABSTRACT

Granulosa tumors are rare ovarian tumors they belong to sex-cord and stromal tumor family. They are the most common type of malignant stromal tumor and have a good prognosis. There are two types: the adult type (AGCTs), which occurs most frequently between the ages of 40 and 70, and the juvenile type (JGCTs), which is uncommon. Juvenile granulosa tumors tend to involve a single ovary and occur mainly in people who are younger than 30 years. Metastatic spread is rare and, if present, is usually limited to the peritoneal cavity. We report a case of ovarian juvenile granulosa cell tumor extending to the jejunum and peritoneum in an 18-year-old female patient received in our institution.

Keywords: Ovarian tumor, Juvenile granulosa cells, Jejunum and peritoneum

INTRODUCTION

Granulosa tumors are rare ovarian tumors which develop in the mesenchyme and sex-cords of the ovary. They represent 0.6 to 3% of all ovarian tumors and 5% of malignant ovarian tumors and are the most common type of malignant stromal tumor.[1] There can be divided into two subtypes according to the differences of the age of patients, clinical and histopathologic: the adult type (AGT), which occurs most frequently between the ages of 40 and 70, and the juvenile type (JGT). TGJ is characterized by a relatively younger age of occurrence, a different morphological aspect with more intense histological signs of malignancy and a higher risk of recurrence.[2-4]

Diagnosis is histological, based essentially on morphological characteristics. The outcome is generally good when diagnosis is made early, but delayed diagnosis is associated with a high risk of peritoneal swelling before and during surgery.[5] We report a case of a juvenile granulosa tumor of late discovery, observed in our facility, which is a level 2 hospital at 702 km from Dakar, the Senegalese capital.[2]

CASE REPORT

This 18-year-old patient was admitted for abdominal and pelvic pain with nausea and vomiting. Physical examination revealed a flexible abdomen and a voluminous, solid, mobile mass covering the right pelvic fossa and hypogastrium. There was no dullness of the flanks or hepatosplenomegaly. Vaginal examination revealed a normal-sized uterus with a right renitent mass mobile and painless on mobilization. The breast examination and the remainder of the physical examination was normal. Abdominal ultrasound finds a heterogeneous myometrium related to multiple

hypoechoic sub centimetric nodules and a heterogeneous adnexal mass 18 cm of large. Computed tomography (CT) scan was not performed. Biological tests showed anemia of 7.1 g/dl, thrombocytosis of 611,000 mm^3, C-reactive protein of 138.3 mg/l and renal failure with creatinemia of 35.4 mg/l.

Surgical exploration by laparotomy found a large tumor of right ovary, friable with some adhesions to the abdominal wall measuring 20 cm long, a solid and obstructive jejunal tumor of 7 cm of diameter, with upstream dilatation and a flattening downstream intestine's, 40 cm from Treitz's angle. the presence of lymphadenopathy, absence of ascites or hepatic nodule (Figure 1).

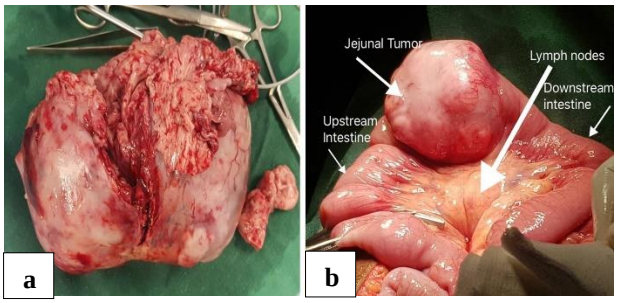

Figure 1: Per operative view (a) ovarian tumor with a reliable appearance, and (b) jejunal tumor with upstream dilatation and a flattening downstream intestine.

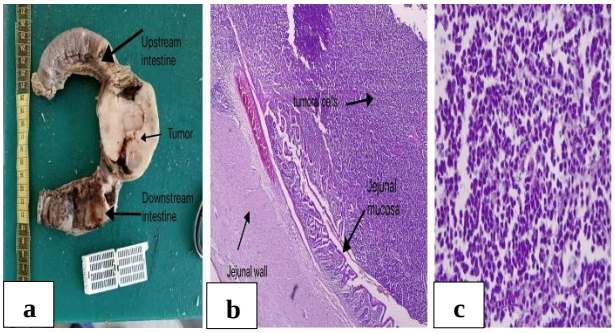

Figure 2: Anatomopathological examination (a) macroscopic appearance on sagittal section; (b) microscopic appearance: tumor invasion of the jejunum, with hematoxylin and eosin (H&E) coloration, magnification x100; and (c) microscopic appearance of the ovarian tumor: tumor proliferation consisting of small, monomorphic cells with abundant, basophilic cytoplasm and an ovoid, vesicular, poorly nucleated fine-chromatin nucleus with rare mitosis. hematoxylin and eosin (H&E) coloration, magnification x40.

A right adnexectomy followed by a bowel resection then a jejunal terminal anastomosis and an omentectomy were performed. The postoperative course was complicated with an occlusive syndrome 10 days after. Surgical revision revealed multiple and voluminous celiac lymph nodes obstructing the duodenum, a loosening of sutures of the jejunal anastomosis. The refection of the anastomosis associated with a gastroenteric-anastomosis was made. The patient had a cardiac arrest which was not recovered in intensive care on day 1 of the repeat operation.

Anatomopathological examination of the surgical specimen revealed a juvenile granulosa cells tumor of the right ovary extended to the jejunum and the peritoneum (Figure 2).

DISCUSSION

There are several histopathological types of ovarian tumors in children. Epithelial tumors which are the most common, germinal cells tumor which derive from the oocyte and tumors from the ovarian stroma and sexual cords. Granulosa tumors are rare ovarian tumors with belong to the family of stromal and sex cord tumors. They represent 5% of ovarian tumors in children and adolescents.[5] The juvenile form tends to affect only one ovary and is diagnosed in patients under 20 years of age in 80% of cases and under ten years of age in 50% of cases, giving it its name.[6,7] Juvenile granulosa cells tumor can be associated with Maffucci syndrome or Ollier disease or isolated as it was in our case. It is then characterized sometimes by a slow and painless progression which can delay the diagnosis in our rural context. The most common symptoms are precocious puberty associated to hypersecretion of estrogen by tumor cells and signs of abdominal overdistention related to an abdominal mass.[8] Imaging can characterize the tumor but cannot easily distinguish to other ovarian neoplasia. On ultrasound and CT scan appear as a solid, unilateral, multi-lobulated tumor with multiple thin or thick and irregular partitions. On magnetic resonance imaging (MRI), these tumors present a T1 hypersignal, related to the hemorrhagic changes. On T2, JGCTs present an intermediate signal, they have a spongy appearance, indicating alternating solid and cystic spaces.[9]

When an ovarian tumor is suspected, the dosage of serum tumor markers such as alpha-feto-protein (AFP), beta human chorionic gonatropin (HCG), cancer antigen 125 (CA125), lactate dehydrogenases (LDH) and inhibin A and B, must be carried out. These markers help guide the diagnosis. In the context of a juvenile granulosa tumor, there is an increase in inhibin A and B because they are secreted in large quantities by the cells. These marks were not available in our context, we performed a laparotomy straight away. Surgery has a double role; on the one hand it allows the staging of the tumor using the international FIGO classification and on the other hand it allows complete excision of the tumor which constitutes the cornerstone of the treatment.[10] It consists on a large excision of the tumor associated with omentectomy. Adjuvant chemotherapy is recommended in advanced and recurrent forms. Currently, the combination of bleomycin, etoposide and cisplatin (BEP) has become the most used protocol.[11] In our patient the tumor was retrospectively classified stage III c (tumor involving one or both ovaries

with peritoneal implants outside the pelvis and/or positive retroperitoneal or inguinal lymph nodes. Tumor macroscopically limited to the pelvis, but with malignant extension to the small or at the omentum, confirmed histologically).

On anatomopathological examination, the macroscopic appearance shows a tumor with a solid component with a multi-cystic appearance, often encapsulated. On section, the tumor appears yellowish or grayish, with areas of hemorrhage and necrosis.[12] In our case, the tumor measured 18 cm in size and the same appearance was found on jejunal tumor. Histologically, the architecture is often lobulated, with signs of luteinization of the cytoplasm. Some nuclear abnormalities such as atypia and high mitotic activity can be observed.[12,13]

The prognosis of JGCTs depends on several clinical (age, tumor size >10 cm, bilateral tumor, rupture) and histopathological factors (nuclear atypia, mitotic index). However, the most important factor is the stage at the time of diagnosis.[5,12] In our rural context, several factors tend to delay the diagnosis and lead to poor outcomes. The survival of stage Ia patients (tumor limited to the ovary) varies depending on the series from 83 to 98%. the median survival for stages III and IV is five years.[5]

CONCLUSION

Granulosa cells tumors are rare ovarian tumors with belong to stromal and sex-cord tumors family. The prognosis is mostly favorable when the diagnosis is early; a diagnostic delay is associated with a high risk of pre- and intraoperative peritoneal spreading.

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