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Surgical Management of Ovarian Mature Cystic Teratoma in a Pediatric Patient: A Case Report

Dr. Daniel Manzano[1], Dr. Pablo. X. Condo Espinel[2*]

[1]Specialist in Pediatric Surgery at the Hospital de Especialidades de Fuerzas Armadas N°1 Quito-Ecuador
[2]Resident Physician of the Pediatric Surgery Department at the Hospital de Especialidades de Fuerzas Armadas N°1 Quito-Ecuador

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***Corresponding author:** Dr. Pablo. X. Condo Espinel
Resident Physician of the Pediatric Surgery Department at the Hospital de Especialidades de Fuerzas Armadas N°1 Quito-Ecuador

Abstract **Case Report**

Mature cystic teratoma is the most common benign ovarian tumor in female children, which requires a timely diagnosis and complementary studies to differentiate and exclude malignant tumors. Its diagnosis is initially clinical and is confirmed by complementary imaging studies such as ultrasound, tomography, or magnetic resonance in the most cases a management with an ovarian sparing surgery either laparoscopic or via laparotomy is recommended.

Keywords: Mature cystic teratoma, female children, benign ovarian tumors in children, ovarian sparing surgery.

INTRODUCTION

In female children around 90% of ovarian tumors are benign (Losty P *et al.*, 2022) and the most frequent ovarian masses are the germ cell tumors represented mainly by the mature teratomas, thus tumors arising from a pathological undifferentiated germ cell and are composed of tissues derived from the three germ layers, those are subclassified into Dermoid cysts (mature cystic teratoma) and Struma ovarii (mature teratoma), both are considered benign (Weinberger V *et al.*, 2018). The clinical signs and symptoms are a palpable abdominal or pelvic mass and abdominal fullness (Péroux E *et al.*, 2015), acute abdominal or pelvic pain appear due a complication with elevation of inflammatory markers and leucocyte levels (Beger B *et al.*, 2020), tumor markers are rarely elevated in mature teratomas, and high levels suggest a malignant tumor.

Ultrasonography is the gold standard modality for diagnosis of mature teratomas. However computed tomography and magnetic resonance are more useful in the diagnosis of a complication (Beger B *et al.*, 2020). Surgical management is recommended for cystic masses that are symptomatic, complex in character, increasing in size over time, enlarge beyond 4 cm, or signs suggestive of malignancy (Puri P *et al.*, 2023). The surgeries are classified as ovary-sparing surgery or oophorectomy, either laparoscopic or via laparotomy (Cong L *et al.*, 2023). In children, the low malignancy rate leads to increased consideration of ovarian sparing surgery as a better option for mature teratomas (Spinelli C *et al.*, 2021).

CLINICAL CASE

A 2-year-old female patient without history of chronic diseases. She presented a decrease of oral intake and sensation of hard abdomen for 2 months. It is accompanied by a sensation of mass in the inferior abdomen, in addition to the persistence of the abdominal mass so, she went to the emergency room. Physical examination shows that vital signs and curves for weight and height are within normal ranges, left abdominal mass of a 10 cm of diameter, painless and mobile on palpation. The rest of physical examination was unremarkable.

Admission laboratory results show mild neutropenia, isolated lymphocytosis, normal c reactive protein and sedimentation rate, basic metabolic panel and hepatic function tests within normal values. Special laboratory results for tumor markers show no significant alterations (Table 1).

Table 1: Laboratory results at admission

Hematology	Tumor Markers
Leukocytes 5.08 / uL	Ca- 125 (36.4 U/ml)
Lymphocytes 69.6%(3.5 / uL)	Ca- 19-9 (9.7 U/ml)

Hematology	Tumor Markers
Neutrophils 24.3% (1.2 /uL)	FSH (5.92 m IU/ml)
Hemoglobin 14.10 mg/dl	Ft4 (1.49 ng/dl)
Platelets 315 / uL	Alpha-Fetoprotein (1.82 ng/ml)
PTT 6.8 s	(B-hCG) (0.1 m IU/ml)
PT 12.6 s	LH (0.1 m IU/ml)

Abdominal ultrasound reported a bulky complex cystic lesion with internal septa that occupies the four quadrants of the abdomen, avascular with a volume of 457 ml, with a fine capsule (Figure 1).

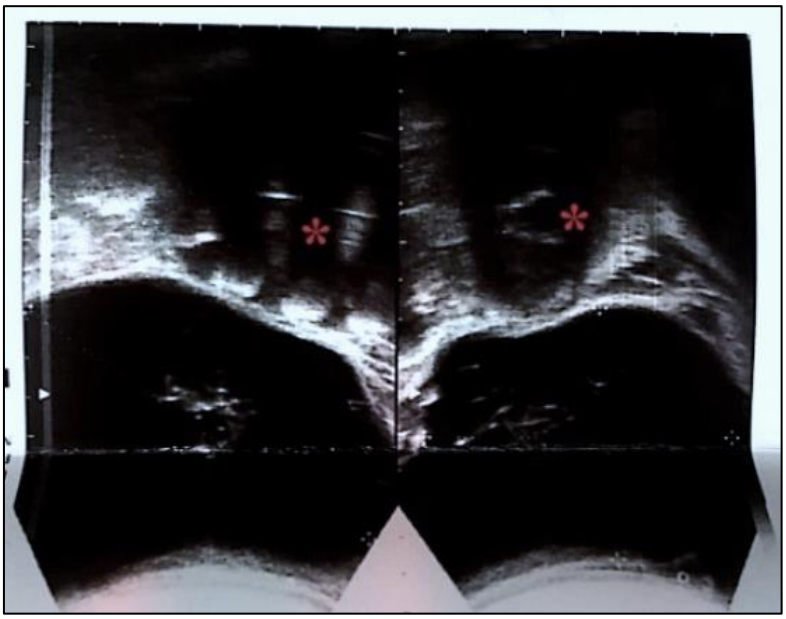

Figure 1: Ovarian cystic lesion with internal septa
Source: Image Service HE1.

Abdominal Computed Tomography without contrast shows a cyst measuring 120 mm x 119 mm x 86 mm with a volume of 638 ml without capsule enhancement (Figures 2, 3 and 4).

Figure 2: Ovarian cystic mass in axial plane
Source: Image Service English Hospital

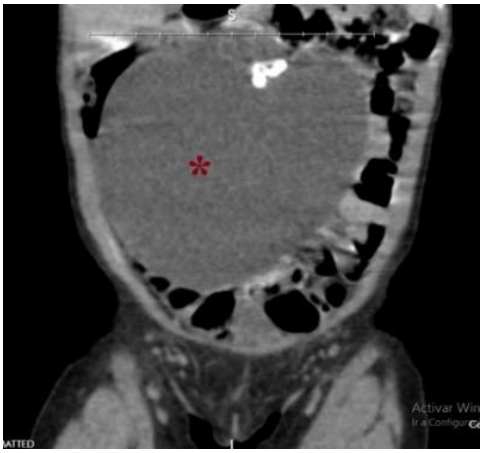

Figure 3: Ovarian cystic mass in coronal plane
Source: Image Service English Hospital

Figure 4: Ovarian cystic mass in sagital plane
Source: Image Service English Hospital

She is admitted for surgical resolution of the mass. Exploratory midline-periumbilical laparotomy was performed with removal of a cystic mass of 13 x 10 centimeters with a volume of 450 milliliters and a weight of 150 grams that was in the left ovary. The right ovary, right fallopian tube and uterus have a good appearance (Figure 5 and 6).

Figure 5: Ovarian cystic mass withdrawal
Source: Pediatric Surgery Service HE1

Figure 6: Ovarian cystic mass
Source: Pediatric Surgery Service HE1

She started intravenous fluids with dextrose 5% and saline isotonic 0.9% at 63 ml/h, analgesic management with intravenous acetaminophen (15 mg/kg/dose) every 8 hours alternated with intravenous ketorolac (10 mg/kg/dose) every 8 hours and intravenous omeprazole (1 mg/kg/dose) each day, she completed 2 days of treatment; She presented adequate oral tolerance to a soft diet 24 hours after surgery, the abdomen was soft, depressed with slight pain at the surgical site, she was discharged 48 hours after the surgery without complications for outpatient control.

The pathological study showed in the macroscopic examination a unilocular cyst, which contains sebaceous material, hair and areas of papillary appearance, hard Rokitansky tubercle is identified (Figure 7).

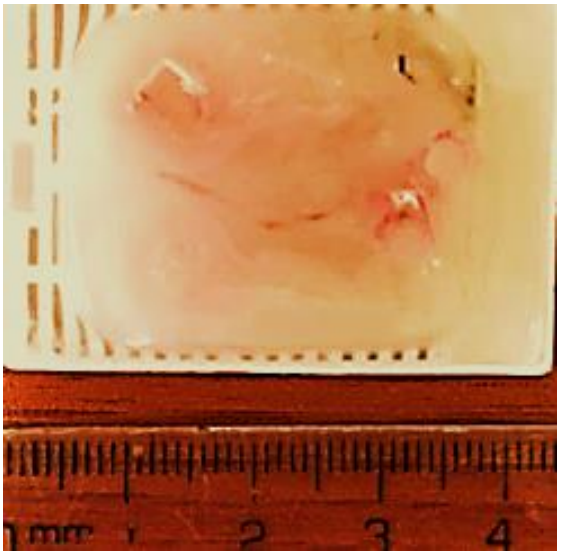

Figure 7: Hair and sebaceous material in the ovarian cystic mass
Source: Pathology Service HE1.

The microscopic examination with hematoxylin-eosin stain showed a cyst that contains hair, bone tissue, cartilage, squamous epithelium, and respiratory tissue in the wall (Figure 8 and 9). The final diagnosis was mature cystic teratoma of the left ovary negative for malignancy.

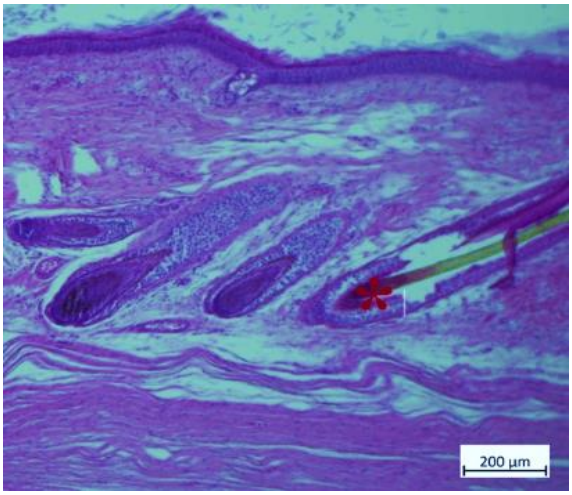

Figure 8: Hair follicle in the ovarian cystic mass
Source: Pathology Service HE1

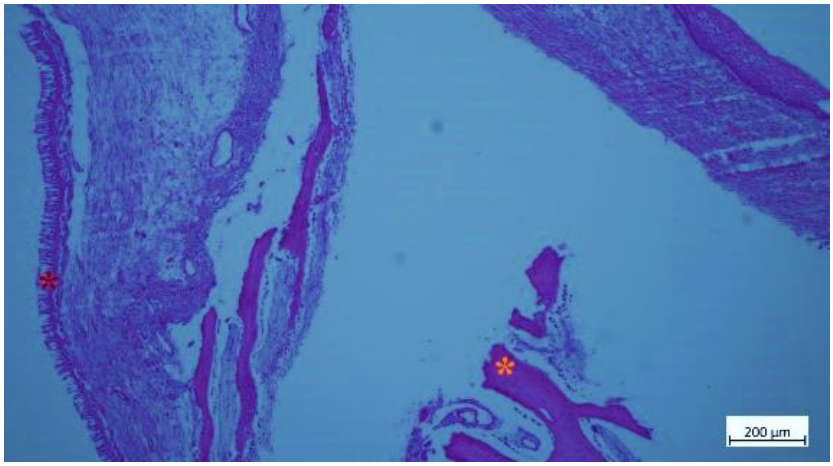

Figure 9: Squamous epithelium and bone tissue
Source: Pathology Service HE1

The patient was evaluated seven days after the surgery, she did not present any symptom or complication. Abdominal ultrasound doing three months later no reported anatomical alterations, ovaries with shape, height and echogenicity inside normal parameters, the patient continue without symptoms (Figure 10).

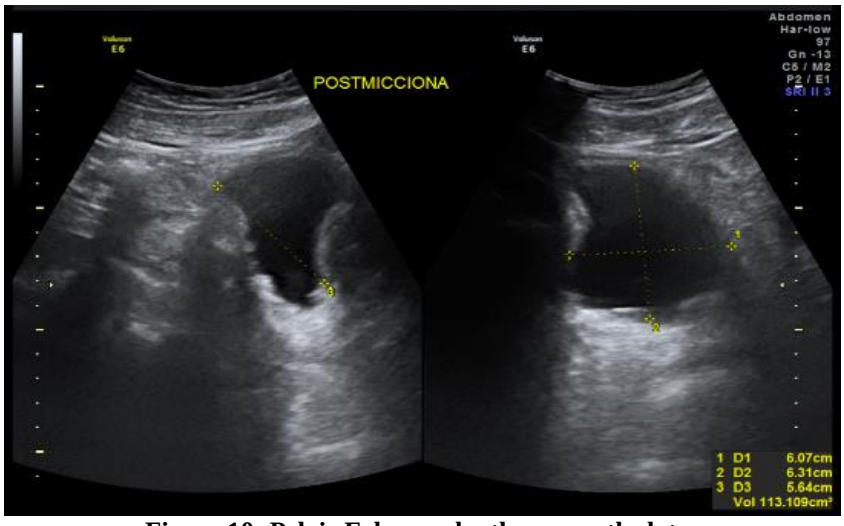

Figure 10: Pelvic Echography three months later
Source: Image Service HE1

DISCUSSION

The annual incidence of ovarian masses in children is approximately 2.6 cases/100,000 girls per year (Prem P *et al.,* 2023), in pediatric and adolescent ovarian masses have a low likelihood of malignancy, approximately 1.5% of all childhood cancers are ovarian in origin, with a malignancy incidence between 0.1 and 1 cases/100,000 girls per year depending on patient age (Renaud E *et al.,* 2019). Around 90% of ovarian tumors are benign, and the proportion of malignancy will increase from birth (18% malignant) until 6 to 7 years old (30% of malignancy) and will decrease drastically thereafter (less than 10% around 14 years old) (Losty P *et al.,* 2022). At the pediatric age the most frequent ovarian histopathological masses are the germ cell tumors meaning 60 – 75% of the cases, represented by the mature cystic teratoma (benign lesion) as the most common histologic subtype of childhood (90% of all cases) (Schneider D *et al,* 2022), the risk of malignancy in mature cystic teratomas is only 2% (Prem P *et al.,* 2023) and around 13% are bilateral (Losty P *et al.,* 2022).

It is thus hypothesized that germ cells tumors, are developmental tumors arising from a pathological undifferentiated germ cell that remains within the gonad since the end of ovarian embryogenesis and, because of ovarian cycle and sequential follicular recruitment, will eventually proliferate and give rise to a tumoral lesion (Losty P *et al.,* 2022). The etiology of ovarian teratomas is unknown and the cell of origin is believed to be totipotent germ cells. Those tumors are composed of tissues derived from the three germ layers (ectoderm, endoderm, and mesoderm). The absence of the immature neuroectodermal component will classify them as mature (the most frequent subtype) or immature (a malignant tumor) (Losty P *et al.,* 2022), the mature ovarian teratomas are subclassified into Dermoid cysts (mature cystic teratoma) and Struma ovarii (mature teratoma), both are considered benign (Weinberger V *et al.,* 2018). The clinical signs and symptoms associated with ovarian cystic teratomas are a palpable abdominal or pelvic mass and abdominal fullness (Péroux E *et al.,* 2015), acute abdominal or pelvic pain due to tumor torsion, hemorrhage, or rupture occurred in less than 16% of the cases and less than 10% are presented with symptoms of hyperthyroidism due to thyroxine production. There are no specific symptoms that indicate if the ovarian mass is malignant or benign (Renaud E *et al.,* 2019). Our patient presented a sensation of mass in the inferior abdomen and signs of abdominal compression causing decrease of oral intake.

Elevation of c reactive protein, CA 19-9 and leucocyte levels may be a marker for dermoid cyst cases with torsion such as a complication (Beger B *et al.,* 2020) but those elements are not presented in our patient. Tumoral markers are frequently used to identify tumor etiology and are rarely elevated in mature cystic teratomas. Alpha-fetoprotein and CA 125 are the most used tumor markers and patients with high levels are more likely to have a malignant tumor and can be useful in differentiating mature teratomas and immature teratomas cases (Luczak J *et al.,* 2018). Thyroid tissue could be a component of the endodermal part of the mature teratoma and can cause increase the levels of free thyroid hormone but in 90% of the patient that not occurred (Renaud E *et al.,* 2019) as observed in the case presented, with no increase levels of tumoral markers or thyroid function test.

Radiologically ultrasonography is the gold standard modality for diagnosis of mature cystic teratoma showing an heterogenous mass with hyperechoic nodule and echogenic shadowing, however computed tomography and magnetic resonance are more specific in the diagnose of pelvic masses with a 87% and 97% respectively, and are more useful in the diagnosis of a complication such as torsion or hemorrhage (Beger B *et al.,* 2020). In the case of our patient an ultrasonography a great cystic lesion with internal septa was identified and an abdominal computed tomography without contrast shows a cystic mass without capsule and no signs of torsion or hemorrhage.

Surgical management is recommended for cystic masses that are symptomatic, complex in character, increasing in size over time or signs suggestive of malignancy (Prem P *et al.,* 2023), ovarian cyst enlarge beyond 4 cm, have an increased risk of torsion, rupture and hemorrhage (Prem P *et al.,* 2023) and have a surgical criteria for intervention. The surgeries were classified as ovary-sparing surgery or oophorectomy, either laparoscopic or via laparotomy (Cong L *et al.,* 2023). Laparoscopic is the gold standard for mature cyst teratoma surgery, reduces the risk of post operative pain, blood loss, and a shorter hospital stay, however increase the risk of intraperitoneal cyst rupture, chemical peritonitis, adhesion formation, iatrogenic spill of malignant cells and high recurrence rates (Luczak J *et al.,* 2018) (Cong L *et al.,* 2023). It is considered that mature cystic teratomas with a diameter around 5 cm or greater in premenopausal women require laparotomy resection, the operative approach can be through a midline laparotomy or Pfannenstiel incision (Prem P *et al.,* 2023) (Cong L *et al.,* 2023). In children, the low malignancy rate leads to increased consideration of ovarian sparing surgery as a better option for mature cystic teratomas and should always be considered in the following cases: no evidence of lymphadenopathy or liver or lung metastasis, normal levels of tumor markers, absence of calcifications, or specific radiological findings (Spinelli C *et al.,* 2021). In our case a midline-periumbilical laparotomy was performed with removal of a cystic mass of 13 x 10 centimeters with preservation of left ovary.

The dermoid cyst (mature cystic teratoma) histopathological contains somatic tissue such as hair, teeth, sebaceous glands from any of the three embryogenic germ layers (endoderm, mesoderm, and ectoderm), the tissue does not contain neuroectoderm

(Kummar B *et al.*, 2018). In our case the pathological pieces contained hair, bone tissue, cartilage, squamous epithelium, and respiratory tissue.

CONCLUSION

We consider that all female children even at a young age (as our case), with a progressive growing abdominal mass associated with a digestive symptomatology or pain, always suspect the possibility of ovarian mass origin, and the need of make complementary studies such as tumoral markers and images, that would allow bring us the majority quantity of information to decide the treatment and if required the surgery process. Due to the size of the mass, the risk of rupture and torsion, and the high probability of a benign process in our patient, the laparotomy with the teratoma excision and ovary-sparing surgery is considered as the first option of surgical management.

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