South Health and Policy. 2025; 4:414
doi: 10.56294/shp2025414

CASE REPORT

Sertoli-Leydig cell tumor in a child and adolescent consultation patient: case report

Tumor ovárico de células de Sertoli-Leydig en paciente de consulta infanto-juvenil: reporte de un caso

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Cite as: Angulo Peraza BM, Fernández Sánchez D, Pacios Dorado JL, Martínez Pérez GM. Sertoli-Leydig cell tumor in a child and adolescent consultation patient. Case presentation. South Health and Policy. 2025; 4:414. https://doi.org/10.56294/shp2025414

Submitted: 18-03-2025 **Revised:** 22-06-2025 **Accepted:** 20-10-2025 **Published:** 21-10-2025

Editor: Dr. Telmo Raúl Aveiro-Róbalo

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ABSTRACT

Introduction: ovarian cancer is the third most common gynecological cancer and the one with the worst prognosis. Among non-epithelial tumors, there are three groups: pure stromal tumors, pure tumors of the sexual cords and mixed tumors, which originate from the sexual cords and stromal cells, with cell tumors belonging to the latter group by Sertoli-Leydig.
Objective: to describe the clinical case of a patient diagnosed with a Sertoli-Leydig cell tumor at the "Dr. Antonio Luaces Iraola" Provincial Teaching Hospital in Ciego de Ávila.
Case presentation: the case of a 14-year-old adolescent was presented, with a history of apparent health, with early onset of sexual relations since the age of 13, who attended the Provincial Outpatient Consultation of Child and Youth Gynecology reporting menstrual delay accompanied by an increase in abdominal volume without other symptoms. The study carried out confirmed the presence of a right adnexal tumor mass, which is why it was decided to perform surgery. The biopsy performed confirmed the diagnosis of Sertoli-Leydig cell tumor.
Conclusions: the case presented constitutes the first and only existing record of its type attended in the province of Ciego de Ávila. The diagnosis of this entity is due to a thorough physical examination and joint assessment by a multidisciplinary team.

Keywords: Adolescent; Gynecology and Obstetrics; Sertoli-Leydig Cell Tumor.

RESUMEN

Introducción: el cáncer de ovario constituye el tercer cáncer ginecológico más frecuente y el que peor pronóstico presenta. Entre los tumores de estirpe no epitelial existen en tres grupos: tumores puros del estroma, tumores puros de los cordones sexuales y tumores mixtos, los cuales se originan de los cordones sexuales y las células del estroma, perteneciendo a este último grupo el tumor de células de Sertoli-Leydig.
Objetivo: describir el caso clínico de una paciente diagnosticada con un tumor de células de Sertoli-Leydig en el Hospital Provincial Docente "Dr. Antonio Luaces Iraola" de Ciego de Ávila.
Presentación del caso: se presentó el caso de una adolescente de 14 años de edad, con antecedentes de salud aparente, con inicio precoz de las relaciones sexuales desde los 13 años, que acude a Consulta Externa Provincial de Ginecología Infanto Juvenil refiriendo retraso menstrual acompañado de aumento de volumen a nivel abdominal sin otra sintomatología. El estudio realizado confirmo la presencia de una masa tumoral

anexial derecha por lo cual se decide realizar intervención quirúrgica, la biopsia realizada confirma el diagnostico de Tumor de células de Sertoli-Leydig.
Conclusiones: el caso presentado constituye el primer y único registro existente de su tipo atendido en la provincia Ciego de Ávila. Al examen físico exhaustivo y la valoración en conjunto de un equipo multidisciplinario se debe el diagnóstico de esta entidad.

Palabras clave: Adolescente; Ginecología y Obstetricia; Tumor de Células de Sertoli-Leydig.

INTRODUCTION

Ovarian cancer is the third most common gynecological cancer and has the worst prognosis. Tumors that are very different from a histological point of view can originate in the ovary, such as non-epithelial tumors, which account for 10 % of ovarian tumors.[1,2,3]

There are two main groups of non-epithelial tumors: germ cell tumors and sex cord-stromal tumors. According to the World Health Organization (WHO), these are classified into three groups: pure stromal tumors, pure sex cord tumors, and mixed tumors, which originate from the sex cords and stromal cells, with Sertoli-Leydig cell tumors belonging to the latter group.[4]

Statistically, this pathology represents less than 0,5 % of all primary ovarian neoplasms. However, it is the most common virilizing tumor and generally appears in women under 30 years of age, although it can affect other age groups.[5]

According to studies conducted at the National Institute of Oncology and Radiobiology (INOR) in Havana between 2009 and 2019, the incidence of this neoplasm in Cuba is estimated to be less than 0,2 %. In this study, 22 patients diagnosed with non-epithelial ovarian tumors were analyzed, among which Sertoli-Leydig cell tumors were the second most common histological type, accounting for 13,6 %.[6]

In Ciego de Ávila, there are no records of patients diagnosed with Sertoli-Leydig cell tumors, which is why the objective is to describe the clinical case of a patient diagnosed with a Sertoli-Leydig cell tumor at the Dr. Antonio Luaces Iraola Provincial Teaching Hospital in Ciego de Avila.

CASE PRESENTATION

A 14-year-old female adolescent, white, with no apparent health history, who began sexual relations at an early age (13) and had an intrauterine device fitted at the age of 13, attended the Provincial Outpatient Clinic for Child and Adolescent Gynecology on March 22, 2023. She reported a delay in her period since February 18, 2023, with symptoms lasting six days, following menarche at age 10 and a menstrual cycle of 4-5/28-30, accompanied by abdominal swelling but no other symptoms.

Physical examination revealed acanthosis nigricans in areas of flexion, mainly the armpits, genital area, and groin region. No hirsutism, polymorphic acne with scarring in the anterior and dorsal regions of the chest. Erythematous-desquamative lesions were present in the midface region, nasolabial folds, and scalp. The lesions consisted of erythematous plaques with irregular edges, covered by greasy scales and yellowish meliceric crusts, with symmetrical distribution. The associated pruritus was mild to moderate. This clinical picture is consistent with seborrheic dermatitis. A globular abdomen is also noted; it is soft and depressible, and an abdominal tumor occupying the hypogastrium and right iliac fossa measuring approximately 15 to 16 cm is palpable, which is not painful on palpation, with no defense or peritoneal reaction.

The gynecological examination reveals stage IV breasts and pubic hair; adult-type vulva with no external lesions of sexually transmitted infections and type 2 labia minora hypertrophy according to Franco's classification. Speculum examination reveals a nulliparous central cervix with grayish-white foul-smelling discharge.

Vaginal examination revealed an anteverted uterus that appeared to be of normal size and consistency; no pain on mobilization of the cervix. Left adnexa not palpable, right adnexa and uterine body; adnexal mass measuring 14 to 15 cm with regular edges, movable, not hard in consistency, no pain on bimanual palpation.

Given these findings, a gynecological Doppler ultrasound is indicated, which reports: Uterus measuring 52 x 31 x 42 mm, endometrium 3,4 mm. Left adnexa: measures 26 x 20 x 19 mm, with 4 peripheral follicles, none dominant. Right adnexa: complex image with internal echoes, measuring 122 x 85 x 102 mm. Doppler study negative (figure 1).

Based on the above, it was decided to admit the patient for further study and treatment. Hematology tests were also indicated, among which alpha-fetoprotein stood out as a tumor marker, with a value of 0,55 Ul/ml (normal value: 0-15 Ul/ml).

A simple and contrast-enhanced abdominal CT scan with 5 mm slices was then performed, showing: Normal-sized uterus with low-positioned Mutiload IUD. A space-occupying lesion with an intensity of 16-35 UH measuring 162 x 74 x 148 mm is noted, which did not capture the contrast injection and which pushes the left ovary, which

is of normal size, intensity, and morphology. No free fluid in the cavity, no adenopathies, and no bone lesions.

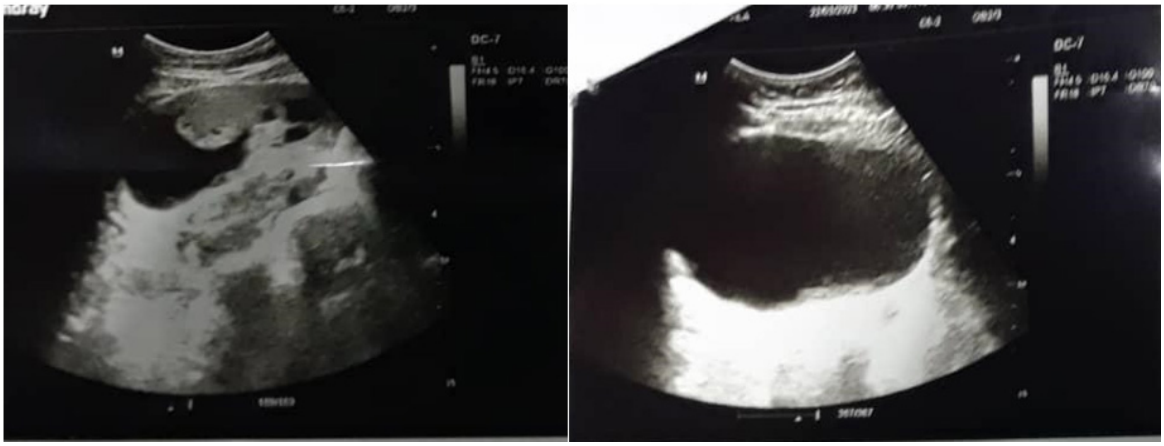

Figure 1. Ultrasound image of the adnexal structures

Once this study was completed, the case was discussed with a multidisciplinary team and an exploratory laparotomy under regional anesthesia was scheduled, which was performed on April 14, 2023.

During the intraoperative period, a uterus of normal size and coloration was observed, with a follicular left adnexa and right adnexa above the uterine body with a tumor mass of approximately 15 cm with a smooth surface and liquid content inside. The rest of the organs in the abdominal cavity were visualized with normal characteristics and no ascites (figure 2).

Figure 2. Image of the tumor taken during the intraoperative period

A total right adnexectomy was performed with wedge resection of the left ovary for biopsy. The intact right adnexa was sent for histological study. At that time, the center did not have the resources to perform a frozen section biopsy, so the specimen was delivered personally to the department of pathological anatomy, which, after performing the study, confirmed the diagnosis of Sertoli-Leydig cell tumor, with intermediate differentiation and a retiform pattern in some areas (figure 3).

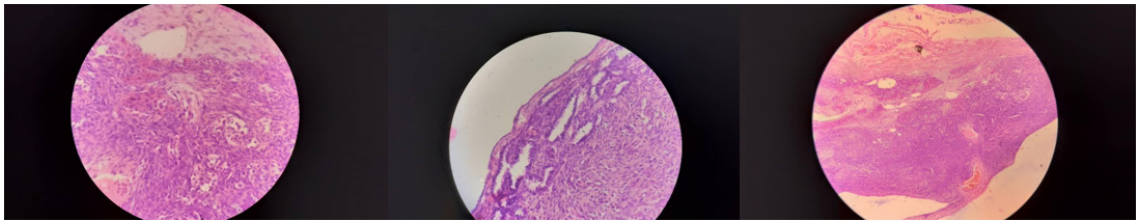

Figure 3. Histological image of the tumor

DISCUSSION

Sertoli-Leydig cell ovarian tumors, also known as androblastoma or arrenoblastoma, generally have low

malignant potential. In most cases, they are well differentiated, slow growing, and diagnosed at an early stage, which gives them a good prognosis. They usually occur between the second and third decades of life, so their discovery in a pediatric patient, as in the case presented, is uncommon.

Although they do not always cause masculinization, most are hormonally active, resulting in clinical virilization, amenorrhea, and hirsutism caused by increased androgen production. Other common symptoms include lower abdominal pain, a feeling of heaviness, and a noticeable increase in volume in that area.[7,8]

Due to its nonspecificity, making a preoperative diagnosis is a real challenge in this type of tumor, and the indication for surgical exploration is based on the criteria for surgery used in all adnexal tumors.[9]

In the case presented, the indication was based on the symptoms (amenorrhea, discomfort, and abdominal distension) and the results of the imaging studies performed. The diagnosis of an androgen-producing tumor was not considered, as there were virtually no elements suggestive of this type of tumor.

Sertoli-Leydig cell tumors are generally characterized by being unilateral, measuring an average of 13 cm in diameter, and typically being solid, yellow ovarian tumors. In this case, the diameter of the removed tumor was 15 cm, with regular edges and a smooth surface.[9,10]

Given these data, surgical management is considered the standard treatment for this type of tumor, with resolution of symptoms and normalization of the hormonal profile. Given the solid characteristics of the tumor, laparotomy has been widely used as the initial surgical approach.[9,11]

As for the prognosis of this type of tumor, the main indicator is the stage of the disease at the time of diagnosis. Fortunately, in most reported cases, the tumor is limited to the ovary, making conservative management possible, which is extremely beneficial in pediatric and adolescent patients, as in the reported case. This is the first and only existing record of its kind diagnosed at the Dr. Antonio Luaces Iraola Provincial Teaching Hospital in Ciego de Ávila, and it was also successfully operated on within the institution. Once the tumor was removed, the patient was able to be transferred in optimal conditions to undergo oncological treatment.[9,11]

CONCLUSIONS

The case presented is the first and only existing record of its kind treated in the pediatric and adolescent gynecology clinic at the Antonio Luaces Iraola Provincial Hospital in Ciego de Ávila. The diagnosis of this condition was made possible by a thorough physical examination and the joint assessment of a multidisciplinary team, since, given that Sertoli-Leydig tumors in young patients generally present with few noticeable symptoms, timely intervention is directly proportional to a better prognosis and reproductive health for the patient, thus contributing to a better quality of life.

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FUNDING

The authors did not receive funding for the development of this research.

CONFLICT OF INTEREST

The authors declare that there is no conflict of interest.

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