From Sertoli-Leydig Cell Tumor to Uterine Inversion and Premature Ovarian Insufficiency

Do Tumor de Células de Sertoli-Leydig à Inversão Uterina e à Insuficiência Ovariana Prematura

Keywords: Leydig Cell Tumor; Ovarian Neoplasms; Uterine Inversion
Palavras-chave: Inversão Uterina; Neoplasias dos Ovários; Tumor de Células de Sertoli-Leydig

Dear Editor,

We report a rare clinical case of Sertoli-Leydig cell tumor (SLCT) culminating in uterine inversion. These tumors represent less than 0.2% of primary ovarian tumors and present diagnostic and therapeutic challenges due to their rarity, heterogeneous clinical presentation, and risk of malignancy.[1-3] The definitive diagnosis is histopathological. First-line treatment is surgical. Adjuvant chemotherapy and radiotherapy may be indicated.[1-4] The prognosis depends on the histological subtype and disease stage.[1-3] Non-puerperal uterine inversion, with multifactorial pathophysiology and morbidity, is a rare gynecological complication.[5] Aggressive behavior and size of the SLCT may contribute to uterine inversion.

A 14-year-old female patient had a history of moderately differentiated SLCT (retiform pattern, FIGO 2021 stage IIB, DICER-1 variant), that was treated three years earlier with right adnexectomy, cytoreductive surgery followed by two courses of adjuvant chemotherapy (bleomycin, etoposide, cisplatin; carboplatin, paclitaxel). She was monitored with MRI and tumor markers biannually for two years, with no recurrence. After this period, she presented abnormal uterine bleeding (AUB) with prolonged and heavy menstruation. The diagnostic tests revealed:

- CA125: 39.2 U/mL (normal < 35 U/mL, suggesting recurrence).
- Transvaginal ultrasound: heterogeneous endometrial thickening, prolapsed into the cervical canal (50 x 46 x 60 mm). Below LO, a cystic formation with solid vegetation (20 x 16 mm).
- Pelvic magnetic resonance imaging: uterus within the vaginal canal with eccentrically located tumor tissue in a horseshoe shape (29 x 65 mm), causing uterine traction. In the mesorectum, there is a solid lesion with 21 mm in diameter. Anterosuperior to the LO, there is another lesion with a diameter of 35 mm.

Given the diagnosis of uterine inversion and persistent AUB with hemodynamic instability, an exploratory laparotomy was performed, with total hysterectomy, left adnexectomy, and excision of a Douglas pouch implant, leaving no macroscopic residual disease (Fig. 1).

The histological diagnosis revealed SLCT with a sarcomatoid component, infiltrating endometrium, myometrium,

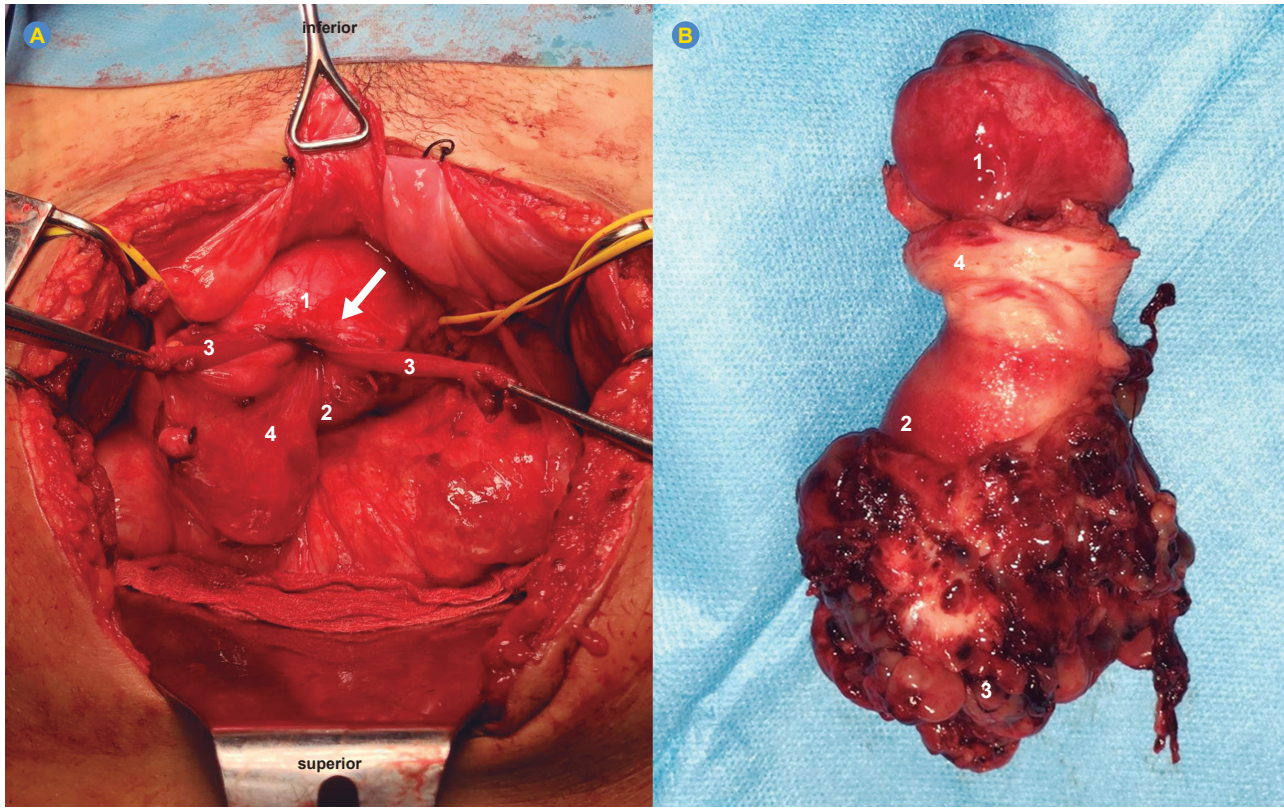

Figure 1 – (A) Surgical findings from an exploratory laparotomy, consistent with uterine inversion, with prolapsed uterus, following prior ureteral marking and bilateral round ligament ligation [(1) anterior uterine wall; (2) posterior uterine wall; (3) bilaterally ligated round ligament; (4) left fallopian tube and ovary; arrow: constriction ring]; (B) Surgical specimen of hysterectomy and left adnexectomy [(1) left adnexa; (2) endometrial epithelium of the uterine body; (3) metastatic endometrial tumor causing inversion of the uterine fundus; (4) cervix].

uterine serosa, endocervix, and left ovarian serosa, without lymphovascular invasion. Following bilateral oophorectomy, the patient met clinical criteria for premature ovarian insufficiency and started hormone replacement therapy. Six months post-surgery, the imaging tests revealed peritoneal recurrence in the pouch of Douglas (two lesions, both 10 mm). Pelvic adjuvant radiotherapy was administered. At sixteen months post-surgery (nine after adjuvant radiotherapy), there were no signs of disease progression.

This case reinforces the importance of early diagnosis and treatment of SLCT, and of referring all patients with SLCT and non-puerperal uterine inversion to specialized multidisciplinary centers.

AUTHOR CONTRIBUTIONS

AM: Data collection, writing of the manuscript.

CSo, CSa: Data collection, critical review of the manuscript.

VS, BL: Critical review of the manuscript.

All authors approved the final version to be published.

PROTECTION OF HUMANS AND ANIMALS

The authors declare that the procedures were followed according to the regulations established by the Clinical Research and Ethics Committee and to the Helsinki Declaration of the World Medical Association updated in October 2024.

DATA CONFIDENTIALITY

The authors declare having followed the protocols in use at their working center regarding patients' data publication.

PARENTAL CONSENT

Obtained.

COMPETING INTERESTS

The authors have declared that no competing interests exist.

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