International Journal of Clinical Obstetrics and Gynaecology

ISSN (P): 2522-6614
ISSN (E): 2522-6622
Indexing: Embase
Impact Factor (RJIF): 6.71

www.gynaecologyjournal.com
2025; 9(6): 116-119
Received: 20-08-2025
Accepted: 25-09-2025

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A rare case of ovarian Sertoli-Leydig cell tumour (Androblastoma) presenting with virilization in an adolescent female

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DOI: https://doi.org/10.33545/gynae.2025.v9.i6b.1735

Abstract
Ovarian Sertoli-Leydig cell tumours are exceptionally uncommon neoplasms in the paediatric and adolescent population, accounting for less than 0.5% of all ovarian malignancies. These tumours typically manifest with clinical manifestations of androgen excess, particularly virilization. This case report describes a 14-year-old adolescent presenting with primary amenorrhoea, progressive virilization, and elevated serum androgens. Comprehensive diagnostic evaluation incorporating hormonal profiling, chromosomal analysis, advanced imaging, and histomorphologic examination revealed a unilateral ovarian Sertoli-Leydig cell tumour of intermediate differentiation. Fertility-sparing surgical intervention via laparoscopic salpingo-ovariotomy was undertaken successfully. Histopathologic confirmation and immunohistochemical phenotyping were consistent with SLCT. Following multidisciplinary tumour board review at the National Cancer Grid, close observation without adjuvant chemotherapy was recommended for this stage IA malignancy. Post-operative surveillance demonstrated normalisation of hormonal parameters, restoration of menses, and clinical remission at two years follow-up without radiologic evidence of recurrence. This case emphasises the diagnostic challenges, fertility preservation principles, and importance of multidisciplinary management strategies in adolescent patients with rare sex cord-stromal ovarian neoplasms.

Keywords: Sertoli-Leydig cell tumour, virilization, adolescent neoplasm, fertility-sparing surgery, sex cord-stromal tumour

Introduction
Ovarian Sertoli-Leydig cell tumours, previously designated androblastomas, represent a distinct subset of sex cord-stromal neoplasms with exceptionally low prevalence in paediatric cohorts. These tumours commonly arise during the reproductive years and characteristically secrete excess androgens, resulting in clinical manifestations of virilization including hirsutism, deepening of voice, clitoromegaly, and menstrual disturbances. Histomorphologic differentiation state and the presence of heterologous components serve as significant prognostic determinants influencing therapeutic decision-making. Given the rarity of these neoplasms in adolescent females, maintaining heightened clinical suspicion remains imperative when evaluating patients with rapid-onset hyperandrogenism and concurrent ovarian pathology, following meticulous exclusion of adrenal and pituitary aetiologies. Comprehensive diagnostic assessment integrating hormonal evaluation, advanced cross-sectional imaging, and definitive histomorphologic confirmation with immunophenotypic characterization remains the cornerstone of accurate diagnosis [1-3].

Case Presentation: A 14-year-old unmarried girl, with no prior therapeutic interventions for her condition, sought clinical evaluation for complaints of absent menarche coupled with progressive development of male-pattern body hair distribution and lowering of voice pitch over a 24-month period. Physical examination demonstrated characteristic features of virilization, encompassing male-type distribution of pubic and abdominal hair, Tanner Stage 3 mammary development, clitoromegaly, and anthropometric measurements within expected range (Figure 1 and 2). Operative-level findings during subsequent surgical intervention revealed a normal-sized anteverted uterus, a marginally enlarged left ovary, and a significantly enlarged right ovary measuring approximately 6 × 6 cm with a glistening white smooth-surfaced mass. Upon sectioning, the tumour demonstrated a red fleshy appearance internally. No macroscopic evidence of peritoneal or omental involvement was identified (Figure 3).

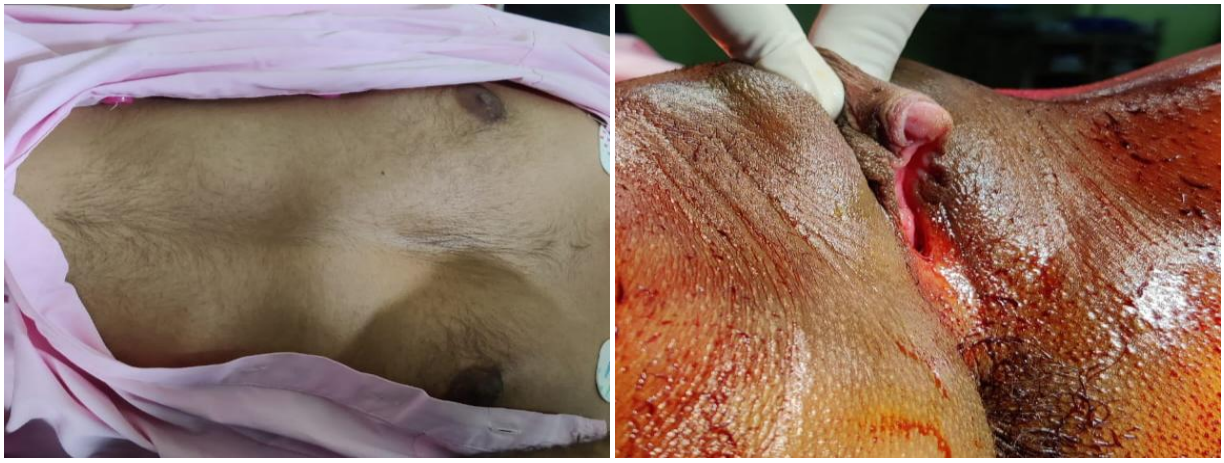


Fig 1-2: Clinical photographs showing male-pattern pubic and abdominal hair with Tanner Stage 3 breast development and features of virilization in a 14-year-old adolescent female.

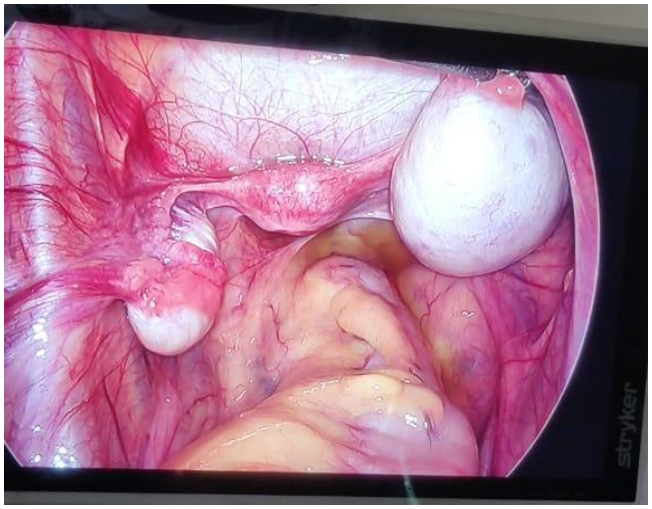
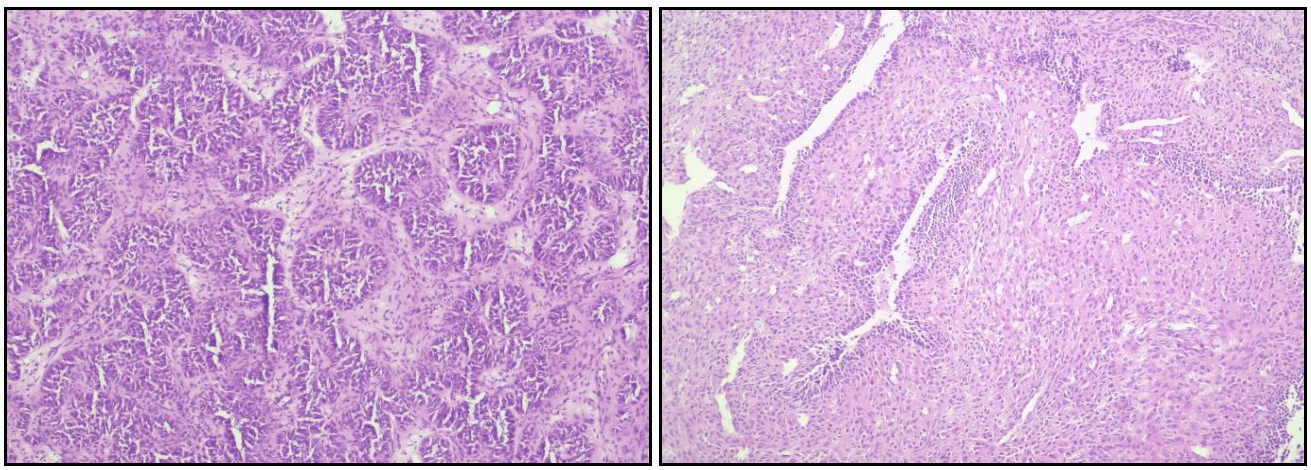


Fig 3: Intraoperative photograph showing the excised right ovarian mass with glistening white, smooth surface and red fleshy mass on cut section.

Investigations

Comprehensive pre-operative laboratory investigation demonstrated normal complete blood count and thyroid hormone parameters. Serum tumour marker assays and sex hormone profiling revealed significantly elevated testosterone concentration with normal dehydroepiandrosterone sulphate levels, indicating an ovarian source of androgen excess. Karyotypic analysis confirmed a normal 46, XX chromosomal complement, excluding chromosomal aberrations. Pelvic magnetic resonance imaging successfully localised a right ovarian mass measuring 37 × 46 × 50 mm with an internal solid component of 40 × 32 × 45 mm. Intraoperative frozen section histopathology was suggestive of Sertoli-Leydig cell tumour. Definitive histomorphologic analysis confirmed an intermediate-grade differentiated SLCT devoid of heterologous elements (Figure 4-7). Immunohistochemical profiling demonstrated strong positivity for inhibin A, Melan A, CD99, WT1, and pancytokeratin, with negative staining for epithelial membrane antigen and CD7, findings consistent with the diagnosis of ovarian Sertoli-Leydig cell tumour.



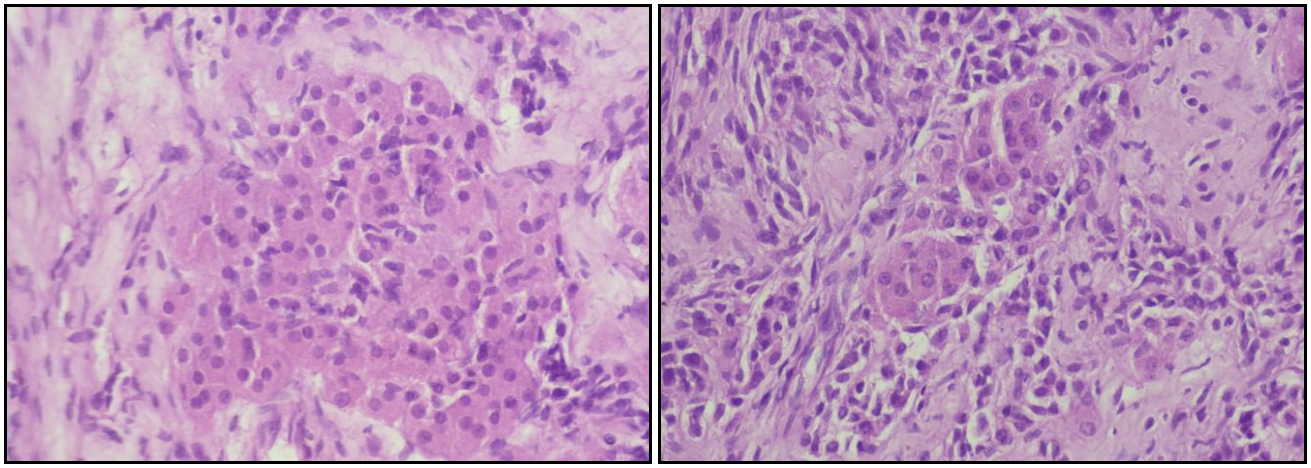


Fig 4-7: Histopathology images at 10X and 40X magnifications demonstrating intermediate differentiation, tumour architecture, and cellular morphology of ovarian Sertoli-Leydig cell tumour.

Differential Diagnosis

The primary differential considerations encompassed polycystic ovary syndrome, which shares certain overlapping endocrine manifestations including hyperandrogenism and menstrual irregularities; however, the presence of a unilateral ovarian mass coupled with markedly elevated testosterone concentration argued against this diagnosis. Adrenocortical neoplasms and bilateral adrenal hyperplasia were entertained in the differential, yet normalised DHEAS levels and radiological evidence of an ovarian mass effectively excluded adrenal pathology. Other sex cord-stromal tumours and androgen-secreting germ cell neoplasms warranted consideration given the clinical presentation of virilization with imaging evidence of an ovarian neoplasm. Additionally, androgen-producing pituitary adenomas, Cushing syndrome, congenital adrenal hyperplasia, and iatrogenic androgen exposure required systematic exclusion through integrated clinical assessment, targeted hormonal studies, and cross-sectional imaging evaluation [2-1].

Treatment

Given the pathologic confinement of the neoplasm to the right ovary with preserved contralateral ovarian tissue, the patient underwent fertility-sparing laparoscopic salpingo-ovariotomy with extraction of the excised specimen within an endoscopic endobag to preclude intra-peritoneal spillage and tumour dissemination.

Outcome and Follow-up

In the immediate post-operative interval, menarche ensued within one month, with subsequent establishment of regular menstrual cyclicity. Structured post-operative surveillance comprised three-monthly transvaginal ultrasonography and serum androgen measurement for the initial 24-month period, transitioning to six-monthly interval assessment for an additional 24 months, followed by annual long-term imaging and biochemical monitoring.

At the two-year post-operative juncture, the patient is expected to demonstrate clinical remission without radiologic evidence of recurrent or metastatic disease [4-5].

Discussion

Sertoli-Leydig cell tumours of the ovary, whilst exceptionally uncommon, manifest predominantly during the reproductive epoch and are fundamentally characterised by pathologic androgen secretion. Contemporary oncologic literature endorses fertility-sparing surgical approaches for stage IA disease, reserving adjuvant chemotherapeutic strategies for histomorphologically poorly differentiated, heterologous-component-bearing, or systemically advanced neoplasms. Multidisciplinary institutional review at the National Cancer Grid Tumor Board by Tata Memorial Centre recommended observational management without adjuvant cytotoxic therapy for this intermediate-differentiated SLCT, an approach substantiated by registry data demonstrating superior oncologic outcomes without chemotherapeutic augmentation in comparable clinical scenarios [6-3, 4, 1].

Systematic diagnostic evaluation in adolescent patients presenting with secondary amenorrhoea and progressive virilization necessitates incorporation of serum androgen quantification. The constellation of markedly elevated testosterone, normalised DHEAS, and imaging evidence of unilateral ovarian pathology directs clinical thinking toward ovarian sex cord-stromal neoplasia, particularly SLCT. Endocrinologic profiling combined with high-resolution cross-sectional imaging are recommended in all suspected presentations, with definitive diagnosis contingent upon immunophenotypic characterisation—notably, inhibin, Melan A, and WT1 demonstrate consistent positive immunoreactivity in SLCT. Contemporary understanding emphasises the prognostic significance of histomorphologic grading and molecular genetic analysis, particularly DICER1 mutational status, in prognostication and treatment stratification [7, 6, 1].

Accumulating evidence substantiates a predominantly indolent natural history for stage IA disease following conservative surgical resection, with documented low recurrence frequencies and favourable extended reproductive sequelae—a pattern mirrored in our patient's clinical trajectory. Malignant biological behaviour correlates with histomorphologic evidence of poor differentiation, retiform growth patterns, or heterologous cellular elements, scenarios warranting consideration of adjuvant chemotherapy. Post-operative surveillance protocols assume heightened importance given documented cases of delayed recurrence and metachronous disease manifestations in this neoplastic subset [3, 5, 4, 6].

Conclusion

This case exemplifies the diagnostic complexity and multifaceted therapeutic considerations inherent in managing rare ovarian sex cord-stromal malignancies in the adolescent population. The successful integration of comprehensive molecular diagnostics, multidisciplinary case discussion, and fertility-sparing surgical methodology resulted in optimal

oncologic and reproductive outcomes. Enhanced clinical awareness regarding atypical presentations of androgen-secreting ovarian neoplasms in paediatric populations, coupled with judicious application of contemporary diagnostic algorithms and evidence-based management principles, remains essential for optimising patient outcomes whilst preserving future reproductive potential.

Ethical Consideration

This case report was prepared following comprehensive review of institutional ethical guidelines. Informed written consent for case publication was obtained from the patient's parents/guardians prior to manuscript preparation.

Conflicts of Interest

The authors declare no competing financial interests or conflicts of interest relevant to this manuscript.

Financial Support and Sponsorship

No specific funding sources supported this case report or its preparation.

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How to Cite This Article

Chaudhary T, Kulkarni S. A rare case of ovarian Sertoli-Leydig cell tumour (Androblastoma) presenting with virilization in an adolescent female. International Journal of Clinical Obstetrics and Gynaecology 2025;9(6):116-119.