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Diagnostic and management challenges in Anti-mullerian Hormone (AMH) secreting sex-cord stromal tumours mimicking Polyendocrine Metabolic Ovarian Syndrome (PMOS) : A case series with review of literature

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Abstract

Background: Polyendocrine metabolic ovarian syndrome (PMOS) is the most common endocrinological disorder among women of reproductive age world-wide. Chronic anovulation, markedly raised serum anti-mullerian hormone (AMH) may be due to small hormone secreting tumours of the ovaries, which are mostly missed due to ovaerlapping features with PMOS. We are presenting the diagnostic and management challenges of anti-Müllerian hormone (AMH)-secreting sex cord stromal tumours (SCSTs) masquerading as polyendocrine metabolic ovary syndrome (PMOS) in three women presenting with chronic anovulatory, infertility and failed response to fertility treatments..

Case presentation: Three reproductive-aged women were referred to our tertiary hospital with presumed PMOS-related anovulatory infertility, markedly elevated AMH levels, and multiple failed ovulation induction cycles. All three women presented with infertility, chronic anovulation, delayed cycles, markedly elevated AMH levels, and multiple failed ovulation inductions. Two had hormonal profiles suggestive of PMOS, while the third had AMH >250 ng/mL, elevated estradiol, and profoundly suppressed gonadotropins (FSH and LH less than 1IU/L). Transvaginal ultrasonography demonstrated a unilateral solid ovarian mass in all cases. Serum inhibin was markedly elevated (>1000 ng/mL), whereas other tumour markers were normal in all the three cases. With provisional diagnosis of AMH secreating sex cord secreting tumours, all three underwent fertility-sparing laparoscopic staging surgery comprising unilateral salpingo-oophorectomy, peritoneal washings, partial omentectomy, and peritoneal biopsies. AMH normalised within one month in all the three. Histopathology confirmed granulosa cell tumour in two patients and Sertoli cell tumour in third. Following tumour removal, both granulosa cell tumour patients resumed menstruation and conceived, resulting in a healthy live birth in one and a biochemical pregnancy in the other. The latter subsequently underwent IVF, yielding nine blastocysts. In the third patient had surgery unmasked premature ovarian insufficiency, necessitating donor-oocyte IVF.

Conclusion: AMH-secreting SCSTs should be considered in women with treatment-resistant PMOS, markedly elevated or rising AMH levels, and a unilateral solid ovarian mass. Early recognition and fertility-sparing surgery are crucial for optimal reproductive and oncological outcomes.

Background

Polyendocrine metabolic ovarian syndrome (PMOS) or Polycystic ovary syndrome (PCOS) is the most common endocrinological disorder of reproductive age women affecting around 8–13% of this population and constitutes as the leading cause of anovulatory infertility. The diagnosis of PMOS is labelled on basis of at least two of three criteria: oligo- or anovulation; clinical and/or biochemical hyperandrogenism; and polycystic ovarian morphology (PCOM), defined as more than 20 follicles per ovary on ultrasound.[1]

Anti-Müllerian hormone (AMH) is a dimeric glycoprotein secreted from the granulosa cells (GCs) and is widely used as a surrogate marker for ovarian reserve for assessing fertility potential, predictability of treatment outcomes and other ovarian disorders affecting ovarian reserve.[2, 3] Being produced mainly by the preantral and small antral follicles in the early follicular phase during follicle-stimulating hormone (FSH) independent stages of development, the expression of AMH decreases in the later FSH-dependent folliculogenesis. [4] As a marker of follicular pool, AMH has been explored to be used as a surrogate marker of PMOS with a high sensitivity and specificity.[5] However, no internationally validated upper cut-off for AMH in the diagnosis of

PCOS has been established due to analytic diversity, missing or contradictory age- and ethnic-specific thresholds, as well as additional factors modulating AMH levels, including obesity, hormonal drug use, and ovarian surgery.

Sex-cord stromal tumours (SCSTs) are rare ovarian neoplasms, comprising of granulosa cell tumours (GCT) and Sertoli-Leydig cell tumours (SLCT) depending upon the active cell type in the malignant tissue.[6] In addition, these tumours are classified according to the hormone being secreted.[7] Granulosa Cell Tumour of the Ovary (GCT) is an uncommon ovarian cancer, accounting for about 5% malignant ovarian cancer.[8] With two sub-types, adult-type and juvenile-type, the adult-subtype is the most common type (95%) and usually develop in perimenopausal or postmenopausal women, at a peak age frequency between 50 and 55 years with few exceptional reports in young adults and children.[9] The juvenile-type GCT is represented in 5% of cases, mostly recognized in the prepubertal age, at a peak age of 13 years.[10] Due to hormone production, most of the cases (97–98%) may show features of hyperestrogenism, such as amenorrhea, dysfunctional menstrual bleeding, growth of uterine leiomyomas, hyperplasia of the endometrium, or endometrial cancer. In rare circumstances, androgen producing GCTs may be present (2–3%) causing virilizing features as secondary amenorrhea, hirsutism, clitoromegaly, deepening of the voice, muscular development and acne due to elevated testosterone levels.[11] Sertoli cell tumours also constitute rare ovarian neoplasms accounting for less than 0.5% of all primary ovarian neoplasms.[12]

There have been few case reports of high AMH in women with sex-cord stromal tumours especially Granulosa cell tumours. This overlap creates an important diagnostic challenge, particularly in women with infertility and are confused with PMOS. A handful of case reports have been published in literature, where in view of high AMH and bulky ovary, misdiagnosis of PMOS has been made, but later on underlying Granulosa cell tumour was recognised. [13, 14] Here, we present three cases in reproductive age group women, who presented with infertility, chronic anovulation and high AMH who were initially labelled as resistant PMOS. On detailed evaluation, all were diagnosed to have AMH secreting sex-cord stomal tumours. Detailed clinical presentation, imaging, tumour markers, surgical procedure, menstrual and reproductive outcomes are presented in this paper.

Case presentation

Three reproductive-age women (aged 18–40 years) who presented with chronic anovulation, infertility, elevated AMH levels and labelled as resistant PMOS were referred to our tertiary referral hospital. Clinical presentation, imaging findings, tumour markers, surgical details and follow-up of three infertile women with presumed diagnosis of PMOS is presented. Ethical approval was taken from the Institute Ethics Committee (IEC) (AIIMSA 6586/05.05.2026) for publishing this case series. Written informed consent was taken from all the patients to use their clinical details for the publication purposes.

Case 1

A 30-year-old P1L1 woman presented with complaints of secondary infertility of four years and irregular menses requiring progesterone therapy for withdrawal bleeding. She had previously been diagnosed with PMOS in view of bulky ovaries (right ovary follicle number per ovary > 50 and left ovary follicle number per ovary

20) at a primary care facility and had undergone multiple cycles of ovulation induction with letrozole alone (up to 5 mg) and along with gonadotrophin based ovarian stimulation cycles without follicular response. With this she was labelled as resistant PMOS and was referred to our centre for laparoscopic ovarian drilling. Detailed history revealed that oligomenorrhea and menses only on progesterone withdrawal were preceded by heavy menstrual bleeding (HMB) for few months. Table 1 shows the details of investigations performed during evaluation at our centre. Her AMH initially one year before was 18 ng/mL and had risen to 48 ng/mL on repeat testing. Inhibin B was markedly elevated at > 1000 pg/mL. General physical examination was unremarkable with no evidence of clinical hyperandrogenemia and her BMI was 19.5 kg/m^2. Transvaginal scan revealed bulky right ovary (74cc) with 3x4cm solid mass with increased vascularity(Fig. 1a and b). MRI confirmed the finding with 5x5 cm heterogeneous solid nodular lesion with mixed T2 signal (Fig. 1c). In the presence of solid ovarian tumour and markedly elevated serum AMH and inhibin B levels, a provisional diagnosis of Sex Cor Stromal Tumour was made and surgery was planned. Fertility sparing staging surgery including laparoscopic right salpingo-oophorectomy with peritoneal washings, partial omentectomy, and multiple peritoneal biopsies was performed. Final histopathology confirmed an adult-type granulosa cell tumour stage IA (Fig. 3 and b). Serum AMH levels normalized within one month of surgery, spontaneous menses resumed and the patient conceived with first cycle of ovulation induction with letrozole (5mg) three months post-operatively followed by normal vaginal delivery of a full term healthy child.

Table 1
Clinical, endocrine and radiological parameters of the cases

Parameters	Case 1	Case 2	Case 3
Age (years)	30	37	29
Presenting complaint	Secondary infertility; chronic anovulation	Secondary infertility; Chronic anovulation	Primary infertility; secondary amenorrhea
BMI (kg/m^2)	19.5	29.5	36.0
Menstrual Patterns	Oligomenorrhea	Oligomenorrhea	Secondary amenorrhea
Infertility	Secondary (4 years)	Secondary (2 years)	Primary (12 years)
Infertility factor labelled	PMOS with chronic anovulation (resistant)	PMOS with chronic anovulation (resistant)	PMOS with chronic anovulation (hypogonadotropic hypogonadism)
Hyperandrogenism	Absent	Absent	Absent
Previous treatment received	Multiple OVI cycles with failed follicular response Referred for laparoscopic ovarian drilling	Multiple OVI cycles with poor follicular response; one failed IVF	No treatment
Investigations			
FSH (mIU/L)	2.4 (Normal)	4.7 (Normal)	0.3 (Reduced)
LH(mIU/L)	11.6 (Elevated)	9.6	0.3 (Reduced)
LH/FSH Ratio	4.8 (Elevated)	2 (Elevated)	1
AMH (ng/ml) preoperative	48	> 25	250
SHBG	134.5	113.5	16.5
Inhibin B (pg/mL)	> 1000	> 1100	1044
Estradiol (pg/mL)		341	874
DHEA-S		190ng/dl	874
CA-125 (U/ml)	11	11	9.1
CA-19.9	45.3	10	3.57
CEA		< 1	6.2
AFP(ng/ml)	4.3	3.42	< 0.2
hCG	< 0.2		< 0.2
Insulin (fasting)	4.5	9.1	29.8

Parameters	Case 1	Case 2	Case 3
Imaging findings			
USG	Right Ovary- 5.5x4.8x5.2cm solid vascular mass ; FNPO-26	Left Ovary- 4x4 cm mixed echogenicity mass	Right ovary- 7.5x6.4 cm complex solid ovarian tumours
Contralateral Ovary	PCOM Left ovary AFC 13	PCOM Right ovary AFC 20	Small and atrophic 1–2 follicles
Histopathology and immunohistochemistry	Granulosa cell tumour, adult type Tumour cells are immunopositive for inhibin, FOXL2, pancytokeratin and calretinin (focal) while they are negative for SALL4 and epithelial membrane antigen (EMA)	Granulosa cell tumour	**Sertoli cell tumour of ovary**
FIGO Stage	IA	IA	IA
Post-operative recovery	Uneventful	Uneventful	Uneventful
Post-operative Course			
AMH(ng/ml)	3.2	0.75	0.02
Inhibin B(pg/ml)	127.7	163.38	5.5
Menses resumption	Yes	Yes	no
Conception	Spontaneous	Biochemical pregnancy	Awaiting donor oocyte IVF

Case 2

A 37-year-old P0 + 0+1 + 0 woman presented with secondary infertility, oligomenorrhea and requirement of progesterone therapy for withdrawal bleeding. She had been treated for several years as a case of resistant PMOS. Her previous treatment history revealed failed response to letrozole and gonadotrophin based ovulation induction cycles. She had undergone one IVF cycle in which only 3 oocytes were retrieved. General physical examination was unremarkable with no features of hyperandrogenemia and her BMI was 29.5 kg/m^2. Transvaginal scan revealed bulky left ovary with a solid mass so was advised for MRI pelvis which demonstrated 5.4 x 4.5 cm solid left ovarian mass along with polycystic morphology in contralateral ovary with an antral follicle count of > 20 (Fig. 1c and d). Her serum AMH level was > 25ng/ml and inhibin B was > 1100 pg/mL. She underwent laparoscopic left salpingo-oophorectomy, peritoneal washing, multiple peritoneal biopsies and partial omentectomy. The specimen retrieval was done in specimen retrieval bag (Fig. 2). Histopathology confirmed an adult-type granulosa cell tumour, stage IA (Fig. 3a and b). Menstrual cycles resumed spontaneously within 2 months post-operatively, and she had a biochemical pregnancy four months after surgery. The patient subsequently underwent IVF in view of advanced maternal age, demonstrating a

markedly improved ovarian response following ovarian tumour removal, with retrieval of 23 oocytes and formation of nine good-quality blastocysts, likely reflecting normalization of the endocrine milieu after surgery.She is waiting for frozen embryo transfer.

Case 3

A 29-year-old woman, married for 12 years, presented with primary infertility and secondary amenorrhoea for one year. She revealed history of heavy menstrual bleeding with Pictorial Blood Assessment Chart (PBAC) score of 480 for four years prior to amenorrhea and had been labelled as PMOS.

Examination revealed BMI of 36kg/m^2 and no evidence of clinical hyperandrogenemia (acne or hirsutism). Abdominal and per vaginal examination revealed an enlarged uterus approximately 12-week size, firm in consistency with a, non-tender firm right adnexal mass measuring around 5×6 cm, with restricted mobility. TVS and MRI showed a right ovarian solid mass measuring 7.5x6.4 cm, small atrophic left ovary with 1–2 follicles and an endometrial thickness of 4 mm along with a 6x6 cm FIGO type 4 anterior wall fibroid. Hormonal evaluation revealed markedly elevated estradiol (874 pg/mL), suppressed FSH and LH (0.3 IU/L), and inhibin B > 1000 pg/mL, consistent with hormonally active ovarian neoplasm along with paradoxical secondary (functional) hypogonadotropic hypogonadism. After multidisciplinary discussion between infertility and gynecologic oncology specialists, the decision for laparoscopic fertility sparing surgery was taken. Laparoscopic right salpingo-oophorectomy, peritoneal washings, partial omentectomy, and peritoneal biopsies was performed. Specimen was removed in an endobag without spillage. Final histopathology confirmed a Sertoli cell tumour. Post-operatively, FSH increased to 36 IU/L one month after surgery, unmasking underlying premature ovarian insufficiency (POI) that had been suppressed by the tumour's hormonal activity. The patient was counselled regarding the implications of POI and referred for donor oocyte IVF.

Discussion and conclusion

The present case series describe an important but under-recognised diagnostic dilemma of AMH-secreting SCSTs which closely mimic PMOS. All three cases had an unusual presentation of anovulation and poor response to ovulation induction therapies along with very high AMH levels and were initially managed as having anovulatory infertility due to PMOS.

Although most of the SCST present with virilising features, infertility and chronic anovulation may be the only presenting symptom leading to misdiagnosis of these rare ovarian tumours as PMOS. Hormonal assessment in these patients provides an important hint towards the final diagnosis. Bulky ovaries with one larger than the other along with high AMH levels should prompt detailed imaging to exclude small AMH-secreting ovarian tumours, which can be easily overlooked despite careful ultrasonography..

Owing to their rarity, non-specific clinical features, and considerable overlap with severe PMOS, these tumours may remain unrecognized for years, resulting in delayed diagnosis and treatment.. None of the three patients exhibited hirsutism or other features of virilisation; menstrual irregularity and poor response to ovulation induction were the only presenting features. Chronic anovulation and/or secondary amenorrhea usually occurs due to increased levels of inhibin B which subsequently inhibits the secretion of pituitary follicle-stimulating

hormone (FSH).[15] Additionally, elevated estradiol levels may also exert negative feedback at hypothalamic-pituitary axis. This leads to suppression of gonadotropins and hence impaired follicular recruitment resulting in chronic anovulation and infertility. This mechanism likely explains the poor response to ovulation agents. Elevated AMH may also affect the appearance of contralateral ovary leading to polycystic ovarian morphology (PCOM). Our first and second case had PCOM of contralateral ovary, while in third case, ovary was small with 1–2 follicles only. All three cases had normalisation of AMH levels within 1 month of surgery. Complete endocrine evaluation is vital in women presenting with PMOS and infertility who fail to respond to standard ovulation induction therapies and when there is discrepancy between different hormonal values especially in presence of unilateral solid ovarian masses.

In report published by Gica C et al, authors reported a case of GCT who presented with secondary amenorrhea along with unilateral ovarian mass. Contrary to our cases, the AMH levels in their index case was 1.86ng/ml. All the previous cases they reviewed, had not mentioned AMH levels.[16] Contrary to this, main feature in our cases was raised AMH levels along with unilateral bulky ovary which led to clinical suspicion in all our cases. Table 2 describes the cases published with high AMH levels who were misdiagnosed as PM OS and were later on detected to harbour sex-cord stromal tumours.

Table 2
Case reports with AMH secreting Sex cord stromal tumours presenting with features of PMOS

Author	Age (yr)	Complaint	Primary diagnosis	Markers	Treatment	Outcome
Gainder et al (2018) (21)		Secondary amenorrhea & infertility. Resistant to OVI	PMOS	AMH > 20 Inh B > 1122	Laparotomy + LSO+ multiple peritoneal, omental biopsies	Conceived with first cycle OVI
Triantafyllidou et al (2018)	32	Secondary infertility & amenorrhea. Resistant to OVI	CC Resistant PMOS	AMH 25.07ng/ml LH 22.8IU/L	Laparoscopic LSO	Spontaneous conception in the next cycle
Yousefi et al (2018) (22)		Secondary infertility		Inhibin B 3000	Laparotomy	
Phyoe-Battaglia et al (2018)		Delayed menses		AMH 36.2 Inh B > 1300	IVF and cryopreservation f/b Lap RSO + PWC + EA	IVF
		Delayed menses		AMH-14.3	Laparoscopic LSO	IVF
Kim et al (2021) (23)	31	Delayed menses Mild hirsutism	PMOS & endometriosis (2x2cm mass in ovary)	AMH- 23.6	Lap RSO + PWC + EA + peritoneal biopsy + partial omentectomy	Spontaneous menses and spontaneous conception
Gica et al (2021) (16)	26	Secondary infertility & amenorrhea	PMOS	LH- 41.9 AMH-1.86 Inhibin-200pg/ml	Laparoscopic right oophorectomy + peritoneal biopsies	Spontaneous conception
Kitajima et al (24)	17	Delayed menses, clitromegaly	PMOS	76.5	Lap SO	Spontaneous menses
Chi et al (25)	35	Delayed menses, infertility	PMOS	AMH-24.1ng/ml	Fertility sparing surgery	Conceived with IVF
Robeva et al (2026) (26)		Secondary amenorrhea		LH- 31.6 AMH- 17.1	Right ovariectomy	Regular menses

First and second cases with HPE diagnosis of Granulosa cell tumour resumed spontaneous menses from next month only and conceived within 3 months of surgery. First case delivered a full-term healthy baby vaginally whereas second case had biochemical pregnancy and subsequently underwent IVF in view of advanced maternal age. She had a markedly improved ovarian response following ovarian tumour removal, with retrieval of 23 oocytes and formation of nine good-quality blastocysts, likely reflecting normalization of the endocrine

milieu after surgery. This emphasizes the hormonal block which got released post-surgery, thus resuming ovulation and increasing the chances of conception. The third case is unusual as it was diagnosed to have PMOS and was being given ovulation induction trials in view of raised AMH levels. But upon presentation, complete endocrine evaluation revealed extremely high AMH, inhibin levels, estrogen levels and with paradoxical hypogonadotropic hypogonadism. This can be explained by excessive estradiol and inhibin secretion from the tumour, which exerted negative feedback on the hypothalamic–pituitary axis, suppressing FSH and LH levels.[17] Further on follow up, her FSH became 36 IU (one month after the surgery) which again emphasized and enlightens about complicated and atypical presentation of these tumours. Her diagnosis changed from PMOS to hypogonadotropic hypogonadism and finally to premature ovarian insufficiency (POI). The case describes a previously unreported presentation of Sertoli-cell ovarian tumour where ovarian failure had been masked by the tumour and its hormones. Although reported rarely, most of the cases of Sertoli cell tumours in reproductive age group have been confused with PCOS in view of features of hyperandrogenemia. [18, 19, 20] Our patient, in contrast, had no clinical features of hyperandrogenemia, but had just amenorrhea and markedly raised AMH levels.

To conclude, AMH-secreting sex cord-stromal tumours are rare but important mimics of PMOS in reproductive-age women presenting with chronic anovulation and infertility. A markedly elevated or rising AMH/Inhibin-B level, poor response to ovulation induction, suppressed gonadotropins along with presence of bulky ovary or solid ovarian lesion should prompt reconsideration of the diagnosis. Careful endocrine assessment and detailed transvaginal imaging or MRI are essential for early detection. Timely fertility-sparing surgery may restore menstrual and reproductive function in selected patients, while optimising oncological outcomes.

Abbreviations

• PMOS
Polyendocrine metabolic ovarian syndrome
• AMH
Anti-mullerian hormone
• FSH
Follicle stimulation hormone
• LH
Luteinising hormone
• SCST
Sex Cord stromal tumours
• SHBG
Sex-hormone binding globulin

Declarations

- **Ethics approval and consent to participate-** yes, institution permission available
- **Consent for publication-** yes, available. Participants have given written informed consent for their personal or clinical details along with any identifying images to be published in this study.

- **Availability of data and materials-** All data is available with corresponding author and will be made available on reasonable request
- **Competing interests-** None
- **Funding-** No funding
- **Authors' contributions-** RM and ST designed the idea. AN, AR, RR, RS helped in collecting the data and completing the follow up if the study participants. SM and RR performed the histopathological examination of the specimen. All the authors have read the final draft of the manuscript.
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Figures

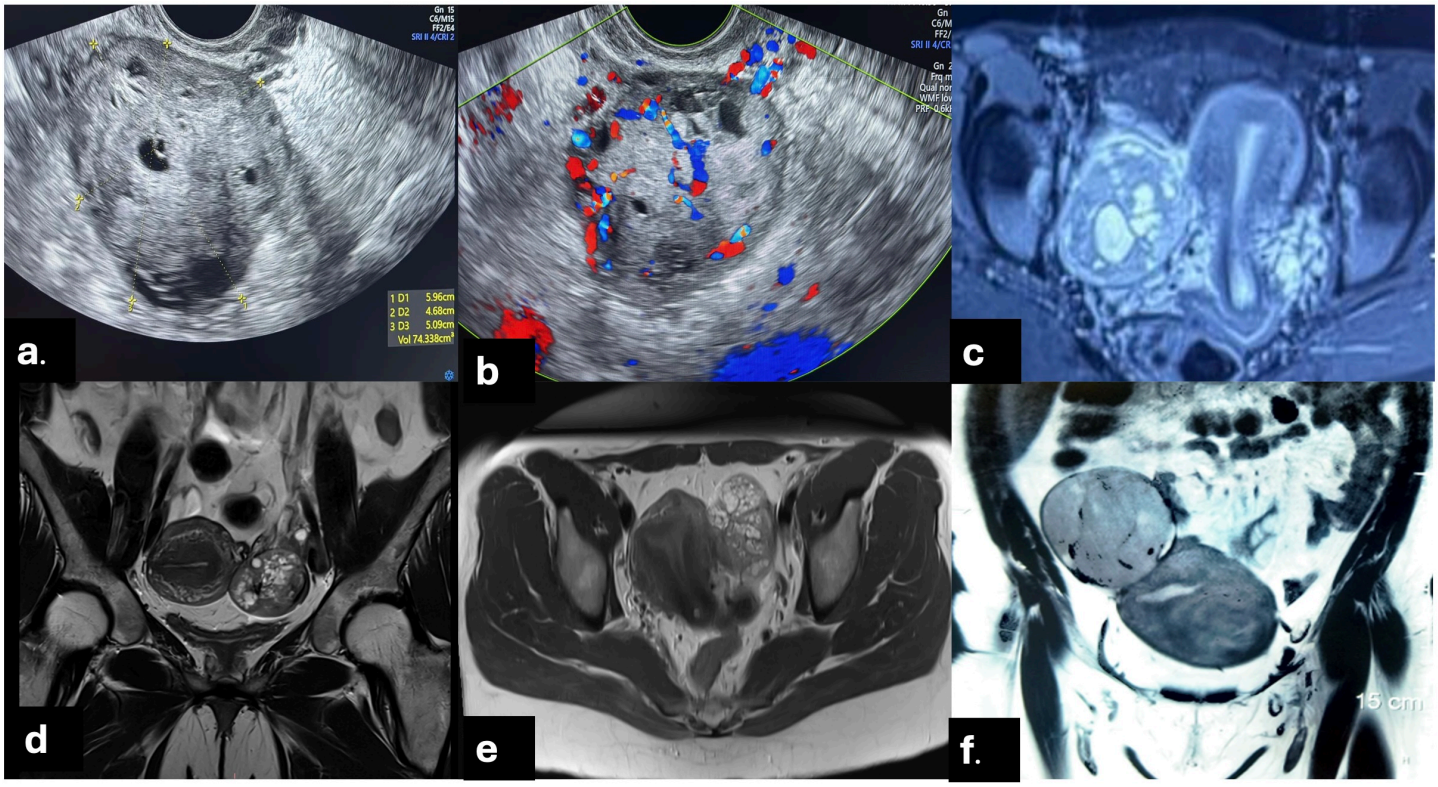


Figure 1

(a and b) -Transvaginal ultrasound imaging of Case 1 with large solid-cystic mass in right ovary with prominent internal vascularity; (c) - Coronal image of Magnetic Resonance imaging (MRI) of same lesion showing heterogeneous solid nodular lesion with mixed T2 signal; (d) & (e) - left solid-cystic ovarian mass in Pelvic MRI of Case 2; (f) - MRI Pelvis image of Case 3 showing complex right ovarian mass.

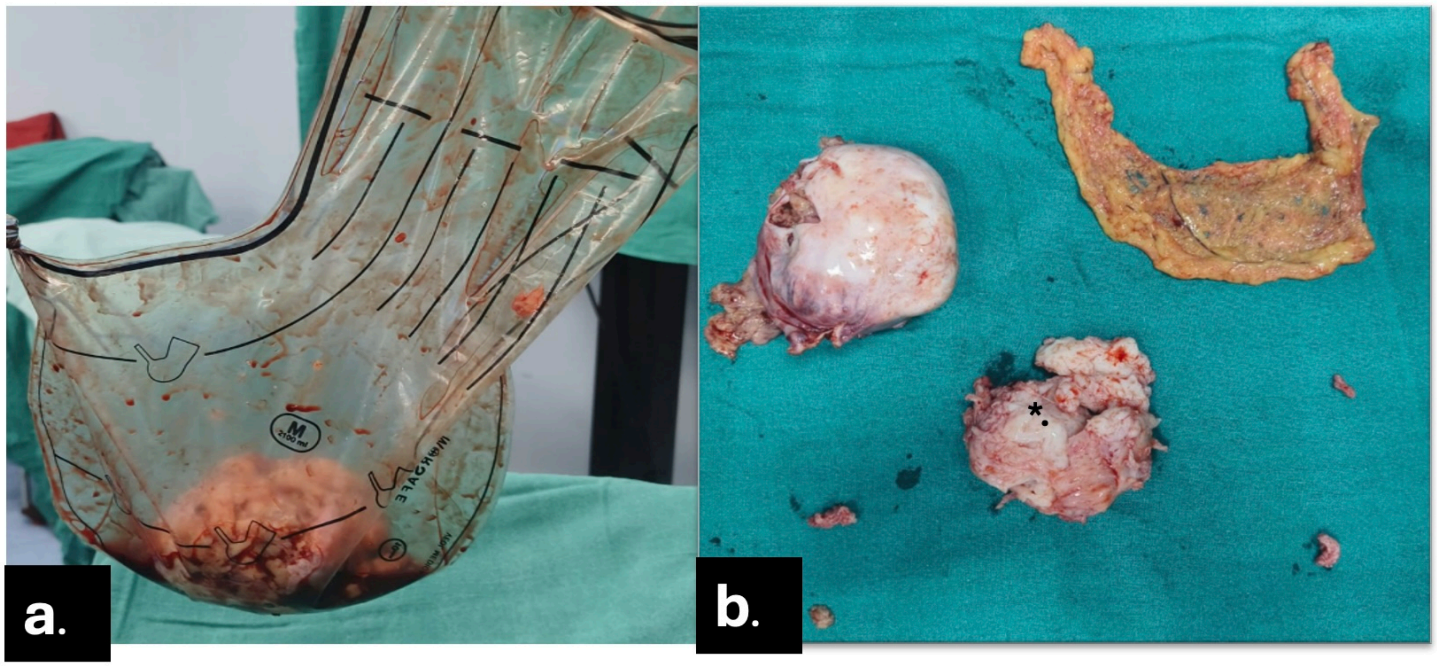


Figure 2

(a) -Laparoscopic specimen retrieval bag (Endobag, medium size, 2100mL capacity) containing the specimen; (b) - gross specimen of smooth white solid ovarian mass, peritoneal biopsies and omentum. On cut-section, the ovary had irregular, friable, cauliflower-like tissue (marked by asterisk) .

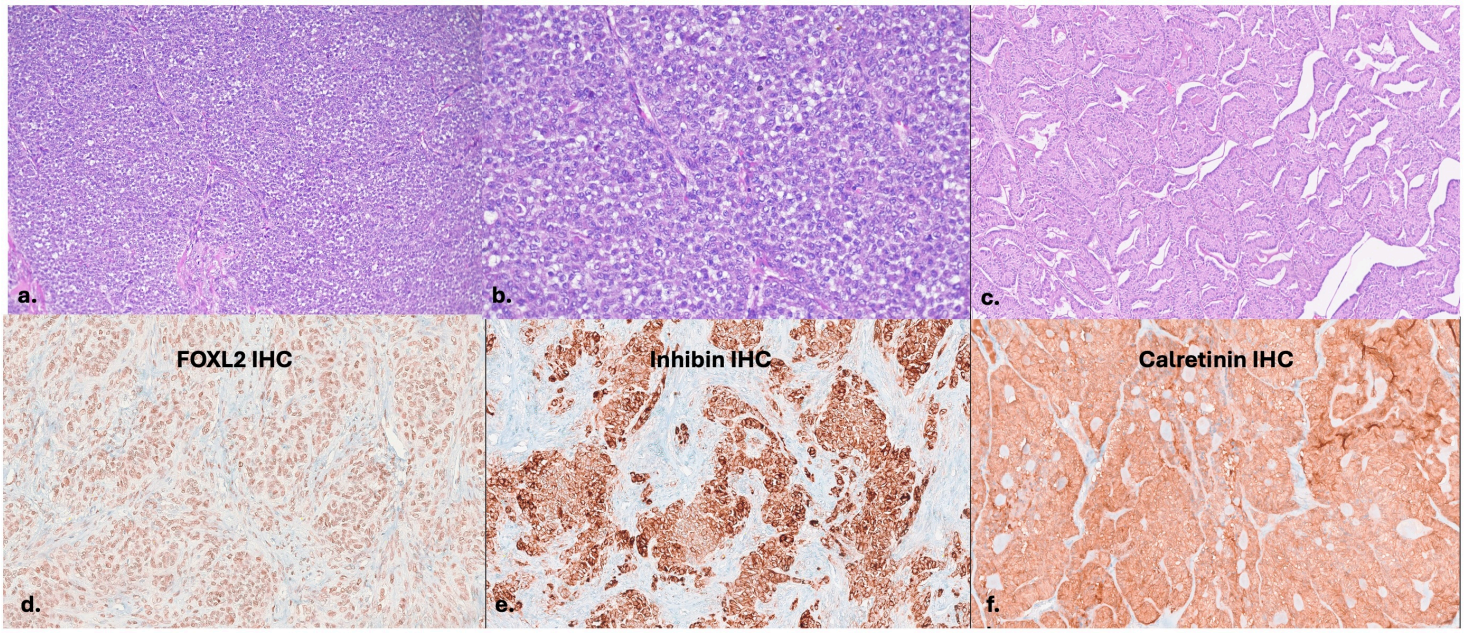


Figure 3

(a) and (b)-microphotograph of adult GCT of ovary (case 1 and 2) demonstrating predominantly diffuse and nodular growth pattern composed of uniform tumour cells with intervening fibrovascular septae with scant to moderate cytoplasm and round to oval nuclei exhibiting nuclear grooves (coffee-bean appearance); (c)- Tumour composed of well-formed, closely packed tubules and cords of uniform Sertoli cells (Case 3). Immunohistochemistry (IHC) and showed diffuse nuclear positivity for FOXL2 (3 d), inhibin (3 e) and calretinin (3 f) supporting sex cord–stromal differentiation