




CASE REPORT OPEN ACCESS

A Rare Case of Large Ovarian Thecoma Diagnosed Histopathologically Following Surgical Management in a Premenopausal Woman: A Case Report and Review of the Current Literature

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ABSTRACT

Ovarian thecomas are rare sex cord-stromal tumors of the ovary. These tumors are estimated to account for 1% of all ovarian neoplasms. Thecomas are primarily composed of cells that resemble theca cells, which are involved in estrogen production. While thecomas are more frequently observed in postmenopausal women, they can also occur in premenopausal women. This report presents a rare case of an ovarian thecoma in a premenopausal woman and reviews the current literature on the topic. Herein we endeavor to contribute to the current body of knowledge and provide guidance to clinicians managing similar cases. Our patient is 43 years old and was diagnosed with an ovarian thecoma after she presented to our facility complaining of abdominal pain and abdominal distension. She underwent a total abdominal hysterectomy and unilateral salpingo-oophorectomy. Histopathological examination confirmed a benign thecoma. We demonstrate that it is crucial to consider this benign condition when evaluating solid ovarian tumors in premenopausal women.

1 | Introduction

Sex cord-stromal tumors (SCST) are a rare form of ovarian neoplasms. Of all forms of SCST, thecomas are the rarest, accounting for only 1% of all ovarian tumors [1]. Ovarian thecomas, or theca cell tumors, are solid stromal tumors made up of lipid-rich cells similar to those of the theca interna, often with variable amounts of fibroblast content [2]. Theca cells secrete estrogen; patients may exhibit signs of hyperoestrogenemia, including irregular

menstrual cycles, endometrial hyperplasia, and endometrial cancer. Thecomas typically present as a unilateral mass and can affect both premenopausal and postmenopausal women. Cases in premenopausal women are rare, with the mean age of onset being 59 years [3, 4]. Imaging is critical in investigating thecomas. The use of a contrast-enhanced computed tomography (CT) scan primarily reveals no enhancement or delayed mild enhancement; an unenhanced CT scan would show the tumor as hypodense or isodense. T1- and T2-weighted magnetic resonance imaging

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Summary

- Ovarian thecomas are benign sex cord-stromal tumors which are unusual in pre-menopausal women.
- These tumors frequently manifest with estrogen-related symptoms such as menstrual disturbances and endometrial changes.
- Diagnosis requires thorough imaging and histopathological analysis.
- Management is largely surgical and for premenopausal patients, fertility-preserving surgery is often effective.
- We highlight here the importance of considering ovarian thecomas when investigating solid ovarian masses.
- By doing so, overtreatment is minimized and patients can be optimally managed.

(MRI) reveal low signal levels of the tumors, which are somewhat enhanced by an MRI contrast agent [5].

The preoperative diagnosis of ovarian thecomas can be challenging due to non-specific presentations. Frequently, these tumors are misdiagnosed as uterine myomas. Surgical management remains the mainstay of treatment; oophorectomy is the standard approach. For those patients who prefer to preserve their fertility, wedge resections can be performed. For postmenopausal patients or those that do not require a fertility-sparing approach, a hysterectomy is the most effective surgical treatment, as ovarian thecomas can present with endometrial hyperplasia or endometrial carcinoma

[6]. Herein, we present a case of ovarian thecoma in a 43-year-old premenopausal patient. The pertinent literature is discussed.

2 | Case History

A 43-year-old premenopausal woman (Para 5 Living 5) presented to our facility with a 2-year history of gradual onset abdominal pain and distension. She reported anorexia, nausea, early satiety with recurrent dyspepsia, increased urinary frequency and urgency, weight loss, and vomiting. On presentation, she was afebrile. She reported no change in her bowel habit, no dysuria, and no abnormal vaginal bleeding or discharge. She had no significant past medical or surgical history. Her obstetric and gynecological history was unremarkable. She had no history of alcohol intake and was a nonsmoker.

On examination, she was pale and afebrile with a blood pressure of 136/86mmHg, a pulse rate of 89 beats per minute, a respiratory rate of 20bpm, and an SpO₂ of 98% in room air. On abdominal examination, her abdomen was grossly distended and revealed a large mass extending to a level corresponding with 38-week gravid uterus. The mass was mobile, smooth, firm, and hard in consistency. On a speculum examination, the cervix appeared normal.

Comprehensive investigations were performed. Laboratory tests highlighted a mild anemia of 12.0g/dL; all other parameters were within normal limits. A cervical smear was negative for intraepithelial neoplasia. Furthermore, an ECHO, ECG, and a chest x-ray were all normal. Cancer antigen 125 (CA-125) was raised at 115U/mL (ref range 0.000–35.000) and carcinoembryonic antigen (CEA) was normal at 1.358ng/mL (ref range

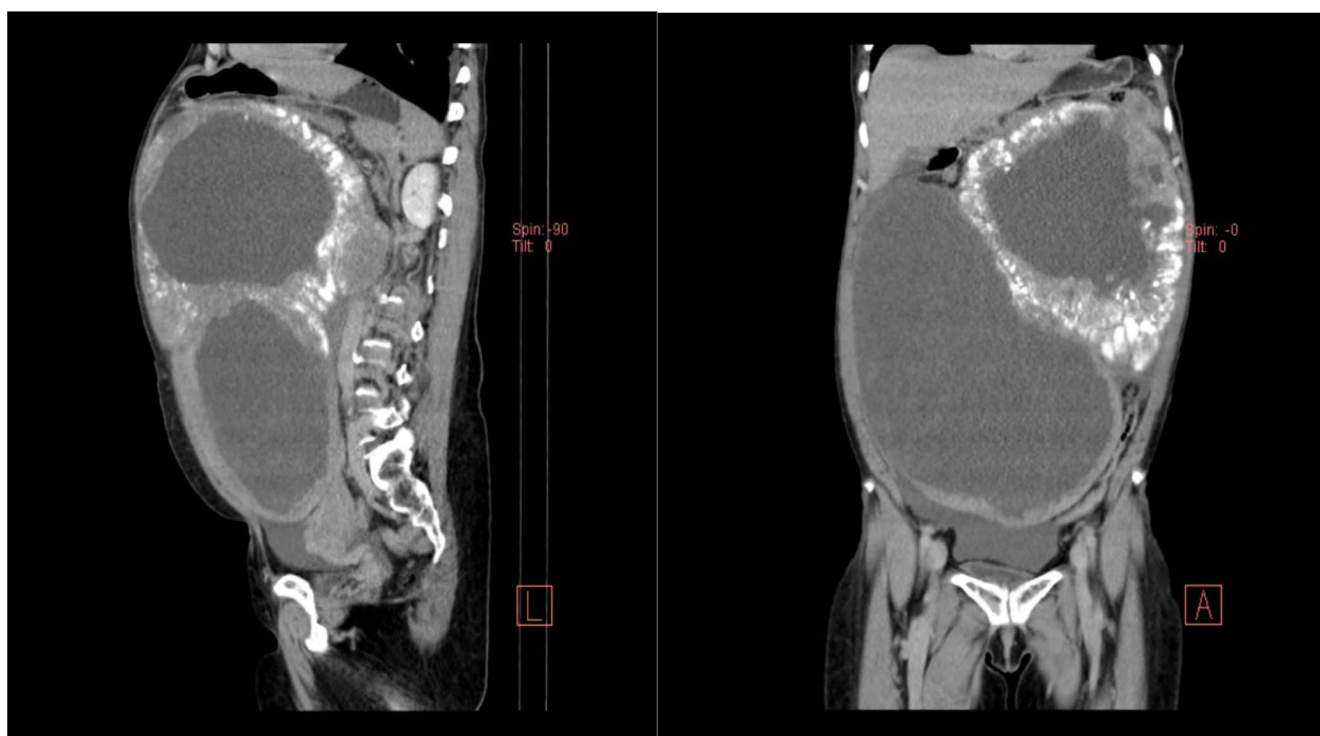


FIGURE 1 | Abdominal CT scan (coronal view) with contrast revealed a large cystic mass with solid component and thick calcified walls. The mass is of unclear origin, displacing abdominal viscera. It measured 27×17×28 cm.

0.00–5.093). β -hCG and α -fetoprotein levels were within normal limits.

A transabdominal ultrasound identified a large immeasurable septated anechoic lesion occupying the pelvic and abdominal cavities; the origin of the lesion could not be identified. An abdominal CT scan with contrast revealed a large cystic mass with solid components and a thick calcified wall. The mass was also of unclear origin and was found to be displacing the abdominal viscera. The mass measured $27 \times 17 \times 28$ cm (Figure 1). There was free fluid in the abdominal and pelvic recess. The differential diagnoses at this time included a complex ovarian cyst, a mucinous cystadenoma, and a serous cystadenoma. The left ovary was not seen.

An explorative laparotomy was performed under general anesthesia, with a left lateral tilt to the operating table. The abdomen was opened in layers with an extended umbilical midline incision. Minimal ascites was present in the peritoneal cavity. The right complex ovarian tumor was identified measuring $23 \times 17 \times 22$ cm, extending up to the xiphisternum. The liver, spleen, and hemidiaphragms appeared normal. A total abdominal hysterectomy and left salpingo-oophorectomy were performed, and hemostasis was achieved. The postoperative recovery was uneventful. Histopathological examination showed a tumor composed of an area with marked hyalinization & collagenisation admixed with regions of plump, ovoid cells with a moderate cytoplasm, coarse chromatic areas, and calcific changes. The histological features were diagnostic for an ovarian thecoma. (Figure 2A–C).

The patient was admitted to the general ward for postoperative care. She received intravenous antibiotics and analgesia. She was discharged on the 4th postoperative day and returned to the clinic 2 weeks later for follow-up with the gynecologist. She will be followed up every 3 months to monitor her progress. To date, regular follow-up evaluation has shown no evidence of tumor recurrence, with negative radiologic findings. Finally, her elevated serum CA-125 level resolved postoperatively.

3 | Discussion

This report discusses a rare case of an ovarian thecoma in a 43-year-old premenopausal patient. This condition is rare globally, and especially in our region of Northern Tanzania. At presentation, our initial clinical impression was an ovarian tumor of unknown type. Following surgical intervention and histopathological assessment, we were able to confirm the diagnosis of an ovarian thecoma. The condition is benign, and so the patient was managed conservatively after surgery. This report explores the rarity of this specific condition, reviews the literature on similar cases, and discusses the management approaches for ovarian thecomas. Thecomas are one form of sex cord-stromal tumors; other examples include granulosa cell tumors and Sertoli cell tumors. Whilst granulosa cell tumors are also considered to be rare entities, thecomas are 1/3 as common as granulosa cell tumors and represent less than 1% of all ovarian tumors [7]. The rarity of this case highlights the importance of building an evidence base for the investigation and treatment of these tumors in the literature.

Furthermore, in patients of this age and menopausal status, ovarian thecomas are an exceedingly rare tumor subtype. Whilst thecomas have been reported in premenopausal and postmenopausal women, the average age of diagnosis is 59 years [3, 4]. These tumors are often estrogen secreting and so their presentation is classically associated with abnormal uterine bleeding, endometrial hyperplasia, and breast pain. It has been reported that 60% of cases present with abnormal uterine bleeding and approximately 20% with concurrent endometrial cancer [8]. Thecomas are also rarely associated with androgenic activity [9]. Of note, our patient was 16 years younger than the average age of presentation and did not report abnormal uterine bleeding. This highlights the need to build an evidence base of the varied spectrum of presentations that can be associated with ovarian thecomas. Whilst abnormal uterine bleeding is a common sign, its absence certainly does not rule out the possibility of an ovarian thecoma. Pelvic or abdominal pain is not a common presentation [9], demonstrating our patient's presentation was unusual.

Ultrasound sonography is the initial imaging investigation. As is common for ovarian neoplasms, thecomas do not have specific features and can manifest as an anechoic, echogenic, or hypoechoic mass on ultrasound [10]. CT is an important investigation, especially in under-resourced facilities such as our own. Thecomas typically manifest as unilateral solid ovarian masses on CT. A common differential diagnosis is a fibroma; MRI can be used to help differentiate between a fibroma and a thecoma. Thecomas are usually hyperintense on T2 weighted imaging, whereas fibromas typically are hypointense. However, the picture is complicated by considering fibrothecomas, whereby the fibroma component can cause hypointensity to be the predominant feature. Reports suggest that thecomas may be accompanied by raised levels of inhibin A and inhibin B, and therefore these laboratory tests could be helpful in raising a clinician's index of suspicion for these tumors [11].

Given the non-specific radiological features of these neoplasms, a histological diagnosis is required. Macroscopically, these tumors are unilateral and small (<5 cm). Stereotypically, these tumors are yellow and solid due to the hormone synthesis that occurs within them. Hemorrhage and necrosis are rare, as these tumors are almost always benign. Plump fusiform cells in nodules or sheets that are arranged in parallel with dense collagen are a histological hallmark of thecomas. Calcification is sometimes seen, as was the case for our patient. Notably, calcific changes are more common in younger patients [7]. The presence of cytoplasmic lipid vacuoles is not required for diagnosis but is not uncommon. Immunohistochemistry can also be used to help inform diagnosis; thecomas have been reported to stain positively for inhibin, calretinin, SF1, and FOXL2 [7]. FOXL2 molecular testing can be used to differentiate thecomas from adult granulosa cell tumors [12]. Molecular testing was not carried out in our patient, highlighting a potential weakness of our case.

The treatment for ovarian thecomas is primarily surgical. The specific treatment is informed by whether the patient requires a fertility-sparing approach. A unilateral oophorectomy is commonly used for a conservative option, whereas a total abdominal hysterectomy is the more definitive treatment option for

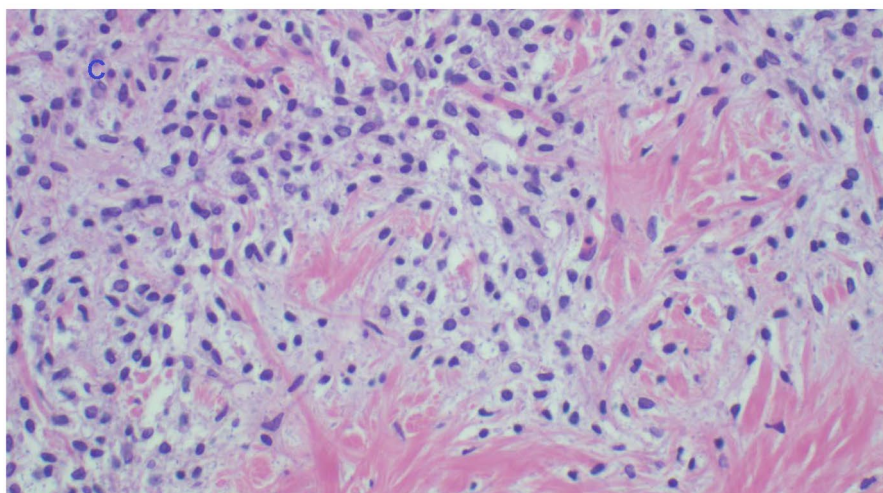
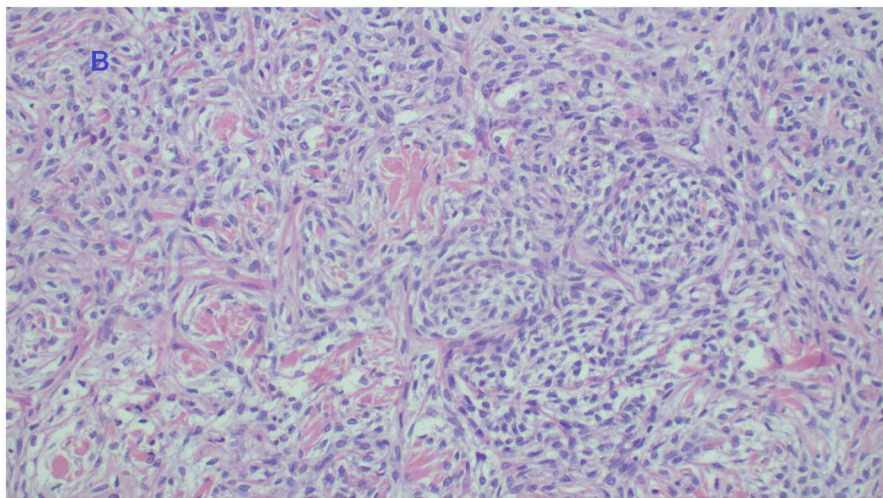
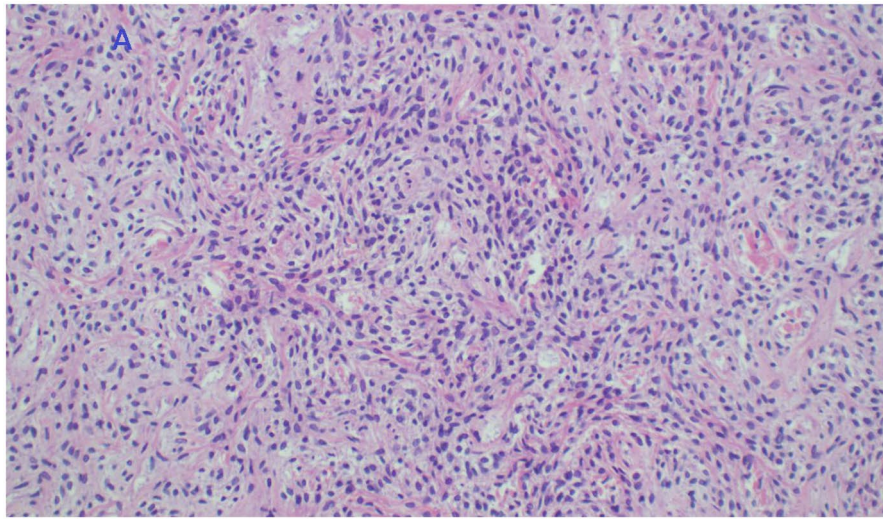


FIGURE 2 | (A) Histopathology of thecoma demonstrating a diffuse growth pattern of ovoid to round cells with pale gray cytoplasm, H&E staining at 40× original magnification. (B) Photomicroscopy of thecoma highlighting hyaline plaques forming keloid-like sclerosis between aggregates of cells, H&E staining at 100× original magnification. (C) Histopathology of ovarian thecoma highlighting areas of abundant hyaline plaques suggesting a thecoma-fibroma hybrid feature; H&E staining at 200× original magnification.

postmenopausal patients or those who have completed their families. As mentioned, 20% of thecomas present with concurrent endometrial carcinomas; these cases are investigated and managed accordingly.

A pre-surgical diagnosis is invaluable, especially for patients who are pre-menopausal and require a fertility-sparing approach. However, this is challenging to achieve due to non-specific symptoms and radiological features. Magnetic resonance imaging is a helpful tool to help diagnose ovarian thecomas pre-operatively [13]. Of note, this is an imaging technique not readily available to patients and clinicians in resource-poor settings such as Tanzania. When pre-operative diagnosis is not possible but frozen section analysis is available, we suggest consenting patients for both fertility-sparing and non-fertility-sparing approaches. If a histopathological diagnosis can be reached intra-operatively, then a fertility-sparing approach can be followed for suitable patients.

Whilst our case refers to a gynecological diagnosis, it has cross-specialty relevance and can be used to inform public health practice. At presentation, our patient was found to have an abdominal mass measuring almost 30cm in length; this suggests that patients are not empowered or able to attend hospital earlier in the disease process. From a public health perspective, a take-away lesson from this case is to use every doctor-patient interaction to remind patients of abnormal symptoms which need to be investigated.

4 | Conclusion

Ovarian thecomas are sex cord stromal tumors of the ovary and make up less than 1% of all ovarian neoplasms. Investigating these neoplasms is challenging, most features are highly non-specific and the diagnosis is rare. It is important therefore to generate a large evidence base in the literature of these cases; this is necessary to identify unifying features from the patient's presentation and investigations that may help clinicians in the future consider the diagnosis of an ovarian thecoma, even when symptoms are atypical. Furthermore, patients with non-specific symptoms often present late in the disease process, patients should be reminded of 'red-flag' symptoms during healthcare encounters.

Author Contributions

John Lugata: conceptualization, data curation, formal analysis, investigation, methodology, validation, writing – original draft, writing – review and editing. **Laetitia Makower:** conceptualization, data curation, formal analysis, investigation, methodology, validation, writing – original draft, writing – review and editing. **Ashley Rapheal:** conceptualization, data curation, formal analysis, investigation, methodology, supervision, writing – original draft, writing – review and editing. **Julius Kaleshu:** conceptualization, data curation, formal analysis, investigation, methodology, validation, visualization, writing – original draft, writing – review and editing. **Tecla Lyamuya:** conceptualization, data curation, formal analysis, investigation, methodology, writing – original draft, writing – review and editing. **Bariki Mchome:** conceptualization, data curation, formal analysis, investigation, methodology, supervision, validation, writing – original draft. **Alex Mremi:**

conceptualization, formal analysis, investigation, methodology, supervision, writing – original draft, writing – review and editing.

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Ethics Statement

The patient provided written informed consent to allow for her de-identified medical information to be used in this publication. A waiver for ethical approval was obtained from the authors' institution review board committee.

Consent

Written informed consent for the publication of clinical details and images was obtained from the patient. A copy of the consent is available for review by the chief editor of this journal.

Conflicts of Interest

The authors declare no conflicts of interest.

Data Availability Statement

No data generated from this study.

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