International Journal of Reproduction, Contraception, Obstetrics and Gynecology
Ramesh B et al. Int J Reprod Contracept Obstet Gynecol. 2026 Nov;15(11):4634-4638
www.ijrcog.org
pISSN 2320-1770 | eISSN 2320-1789

DOI: https://dx.doi.org/10.18203/2320-1770.ijrcog20264024

Case Report

A case report of metastatic ovarian cancer presenting as acute abdomen with obstructed umbilical hernia

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Received: 28 August 2026
Revised: 08 October 2026
Accepted: 09 October 2026

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ABSTRACT

Sertoli-Leydig cell tumours (SLCTs) are rare ovarian sex cord-stromal neoplasms, accounting for less than 0.5% of ovarian tumours. They predominantly occur in young women and are frequently associated with androgenic manifestations. Advanced-stage disease in elderly postmenopausal women is exceptionally uncommon. Here , we present a 76-year-old postmenopausal woman with acute abdominal pain and vomiting due to an obstructed, irreducible umbilical hernia. Emergency laparotomy revealed a friable omental mass within the hernial sac, approximately 2 L of ascites and a large 15×15 cm left ovarian mass densely adherent to the rectosigmoid, bladder, uterus, pelvic sidewall and abdominal wall, with multiple peritoneal tumour deposits. Frozen section showed a poorly differentiated malignant neoplasm, with carcinosarcoma considered. Comprehensive cytoreductive surgery including total abdominal hysterectomy, bilateral salpingo-oophorectomy, peritonectomy, omentectomy, rectosigmoid resection and lymph-node assessment was performed. Definitive histopathology demonstrated a poorly differentiated (Grade 3) Sertoli-Leydig cell tumour with heterologous mucinous and cartilaginous differentiation. The tumour showed surface involvement and rupture, parametrial involvement and extrapelvic peritoneal deposits, with negative regional lymph nodes. No virilising features were documented. This case highlights an exceptionally rare presentation of advanced poorly differentiated SLCT in a postmenopausal woman, masquerading as an acute surgical abdomen due to an obstructed umbilical hernia. SLCT should be considered in the differential diagnosis of ovarian malignancy even in elderly women without androgenic manifestations. Heterologous differentiation may mimic epithelial ovarian carcinoma or carcinosarcoma on frozen section, underscoring the importance of definitive histopathological and immunohistochemical evaluation.

Keywords: Sertoli leydig cell tumour, Ovarian sex cord stromal tumour, Heterologous differentiation, Postmenopause, Obstructed umbilical hernia

INTRODUCTION

Ovarian cancer is one of the major causes of morbidity and mortality among gynaecological malignancies worldwide. It is frequently diagnosed at an advanced stage because of its vague and nonspecific symptoms and the absence of an effective population-based screening strategy. Common symptoms include abdominal bloating, abdominal or pelvic pain, increased urinary frequency, early satiety and altered bowel habits. Hormone-producing ovarian tumours may additionally present with oestrogenic or androgenic manifestations, potentially facilitating earlier diagnosis.[1-3] These may include hirsutism, acne, oligomenorrhoea, amenorrhoea, male-pattern hair loss, clitoromegaly and deepening of the voice.[1,2] However, hormonal manifestations are not universal. In a series specifically examining SLCTs in postmenopausal women,

hyperandrogenism was present in only a subset of patients.[2]

Risk factors for epithelial ovarian cancer include increasing age, family history, nulliparity, obesity, smoking, endometriosis and prolonged exposure to certain hormonal factors. Factors associated with reduced risk include combined oral contraceptive use, tubal ligation or salpingectomy, breastfeeding and multiparity.[1,2]

The pattern of spread differs according to histological subtype. Epithelial ovarian carcinomas most commonly disseminate through the transcoelomic route, with involvement of the peritoneum, omentum and visceral surfaces, followed by lymphatic and direct extension. Sex cord-stromal tumours have distinct biological behaviour and may disseminate through surface extension and haematogenous routes.[2-5]

Sertoli-Leydig cell tumours (SLCTs) are rare ovarian sex cord-stromal tumours, accounting for approximately 0.5% or less of ovarian neoplasms. They predominantly occur in young women, usually during the second and third decades of life, and commonly produce androgens. Fewer than 10% of reported patients are older than 50 years.[1,5-7]

We present an exceptionally rare case of a 76-year-old postmenopausal woman with a poorly differentiated Sertoli-Leydig cell tumour with heterologous differentiation, presenting acutely with an obstructed umbilical hernia. The pelvic tumour was discovered incidentally during evaluation of the acute abdomen and was subsequently found to represent advanced metastatic ovarian malignancy.

CASE REPORT

A 76-year-old postmenopausal woman, para 1 living 1, with a previous normal vaginal delivery, presented to our hospital with acute abdominal pain and vomiting. She had a history of a congenital umbilical hernia that had progressively increased in size and had become irreducible over several months, associated with mild intermittent pain. There was an acute exacerbation of pain associated with vomiting for one day prior to admission.

On abdominal examination, an irreducible, tender umbilical hernia was noted. The abdomen was associated with guarding and rigidity. A provisional diagnosis of obstructed umbilical hernia with possible bowel compromise was made. Initial investigations revealed a haemoglobin level of 11.7 g/dL, normal complete blood count, markedly elevated C-reactive protein of 151 mg/L and serum albumin of 2.4 g/dL. Tumour markers showed CA-125 of 347 U/mL and AFP of 3.4 ng/ml. Contrast-enhanced CT of the abdomen and pelvis performed during evaluation of the acute abdomen demonstrated the obstructed umbilical hernia as well as an incidental large pelvic mass (Figure 1).



Figure 1: CECT whole abdomen and pelvis.
Umbilical hernia ~3 cm with small bowel loops as content. Small mesenteric lymph node, solid. Heterologous mass 12.5x 13 cm, solid cystic component with small fleck of calcification. Uterus & ovaries not separetely visualised. Adhesions to rectosigmoid& small bowel.

In view of the acute presentation and suspected obstructed hernia, the patient underwent emergency exploratory laparotomy under general anaesthesia.The hernial sac was carefully separated from the umbilicus and underlying structures (Figure 2). The sac contained a friable omental mass measuring approximately 5 × 5 cm with multiple nodules suspicious for metastatic deposits (Figure 3). The mass was excised and sent for histopathological evaluation. The incision was extended caudally through a vertical midline approach. Approximately 2 L of ascitic fluid was evacuated and sent for cytological examination.



Figure 2: Umbilical hernia being separated from underlying structures.



Figure 3: Friable omental mass with tumour deposits seen as hernial content.

A large, friable pelvic mass measuring approximately 15×15 cm was identified arising from the left ovary. It was densely adherent to the rectosigmoid colon, urinary bladder, uterus, pouch of Douglas, small bowel, lateral pelvic wall and anterior abdominal wall. Multiple free-floating tumour masses were noted within the ascitic fluid. Hand-assisted adhesiolysis was performed to mobilise the pelvic tumour. The pelvic mass and hernial mass were sent for frozen-section examination. Frozen section revealed a poorly differentiated malignant neoplasm, with carcinosarcoma considered in the differential diagnosis. In view of the extensive disease and the patient's postmenopausal status, the procedure was proceeded with as comprehensive cytoreductive surgery. Total abdominal hysterectomy with bilateral salpingo-oophorectomy was performed. Extensive peritonectomy was carried out around the pelvic tumour. Both ureters were identified and dissected, and parametrial tumour deposits were excised.

The pelvic tumour was densely adherent to the lateral pelvic wall. The specimen was removed En bloc. Approximately 15 cm of rectosigmoid colon, which was densely plastered to the tumour, was resected, followed by two-layer end-to-end colorectal anastomosis using PDS sutures. The integrity of the anastomosis was confirmed intraoperatively. An infracolic omentectomy was subsequently performed. Surgical staging included bilateral pelvic lymph-node dissection involving the external iliac, internal iliac and obturator groups, along with para-aortic and paracaval lymph-node sampling/dissection. The upper abdomen was inspected carefully. No gross metastatic deposits were identified over the liver, stomach or diaphragm. The entire specimen was sent for definitive histopathological examination. The initial intraoperative clinical impression was advanced carcinoma of the left ovary. Microscopic examination revealed a malignant ovarian neoplasm composed of markedly pleomorphic cells with an increased nuclear-to-cytoplasmic ratio, hyperchromatic nuclei, scant cytoplasm and numerous atypical mitotic figures (>20/HPF). Areas of mucinous and cartilaginous differentiation were identified, together with necrosis and calcification.

The final histopathological diagnosis showed Sertoli-Leydig cell tumour of the ovary with heterologous differentiation, poorly differentiated (Grade 3). Additional pathological findings were tumour involving one ovary with surface involvement and rupture. Contralateral ovary was uninvolved. Uterus was unremarkable and intact. One parametrium was involved. Fallopian tubes and cervix were uninvolved. Ascitic fluid was negative for malignant cells. Multiple grey white masses floating in the ascitic fluid was positive for tumour. Regional lymph nodes were negative for metastatic disease. Resected bowel showed no tumour infiltration; areas of luminal constriction were noted. Largest extrapelvic peritoneal tumour focus measured 1 × 0.6 cm. Tumour deposits were identified in the bowel mesentery. Distant metastasis could not be determined. Pathological classification of pT3b Nx Mx was made.

The pathological features of heterologous SLCT are well recognised and may include mucinous/gastrointestinal-type epithelium, cartilage and other sarcomatous elements. These components can make diagnosis particularly difficult, especially in poorly differentiated tumours, and may result in confusion with epithelial malignancies or carcinosarcoma on frozen section.[8,9]

DISCUSSION

Sertoli-Leydig cell tumours are among the rarest ovarian sex cord-stromal tumours, representing approximately 0.5% or less of ovarian neoplasms. They are characterised by variable proportions of Sertoli cells and Leydig cells and are typically unilateral. Most patients are young, with the peak incidence occurring in the second and third decades of life. The classical clinical presentation of SLCT includes abdominal pain or distension associated with androgenic manifestations. However, hormonal manifestations are not universal. Therefore, the absence of virilisation does not exclude SLCT.

Most of the SLCTs present at early stage. Poorly differentiated tumours and tumours containing heterologous elements have a more aggressive clinical course and a greater risk of recurrence.[8-9] SLCTs are classified according to their degree of differentiation into well differentiated, moderately differentiated and poorly differentiated forms. Retiform and heterologous variants are also recognised.[8,9] Poorly differentiated tumours are characterised by immature Sertoli cell structures, prominent atypia and increased mitotic activity. Heterologous elements may include mucinous epithelium,

cartilage, skeletal muscle and other mesenchymal elements.[8,9]

This heterologous differentiation can create substantial diagnostic difficulty. In the present case, the frozen section was interpreted as a poorly differentiated malignant neoplasm, with carcinosarcoma considered. The final diagnosis of poorly differentiated SLCT with heterologous differentiation was established only on definitive histopathological examination. This phenomenon has been described previously, with poorly differentiated SLCTs occasionally being mistaken for epithelial ovarian carcinoma or other high-grade ovarian malignancies on frozen section.[10]

The present case is unusual because of the patient's age of 76 years, advanced disease, absence of classical virilisation and unusual acute presentation. The most striking feature of this case was the presentation as an obstructed umbilical hernia. Umbilical hernias may rarely contain intra-abdominal malignant disease, including ovarian malignancy and peritoneal metastatic deposits. Karabulut et al reported a giant ovarian tumour presenting within an incarcerated umbilical hernia.[11] Similarly, Millar et al described a series of patients in whom ovarian carcinoma presented as an umbilical hernia, with malignant ascites and ovarian malignancy identified at exploration.[12] A further case report described primary peritoneal adenocarcinoma presenting as the content of an incarcerated umbilical hernia.[13] These reports highlight the importance of considering an underlying intra-abdominal malignancy in elderly patients presenting with an atypical, enlarging or newly symptomatic umbilical hernia, particularly when associated with ascites, weight loss, abdominal distension or constitutional symptoms. When an androgen-secreting ovarian tumour is suspected, serum testosterone and DHEAS can help establish the source of androgen excess and distinguish ovarian from adrenal causes.[7,14] In the present patient, no clinical features of virilisation were documented, and the diagnosis was not suspected preoperatively.

Tumour differentiation, stage, mitotic activity, tumour rupture and the presence of heterologous elements are important prognostic factors. Well-differentiated SLCTs generally have an excellent prognosis, whereas poorly differentiated tumours and those containing heterologous elements carry a higher risk of recurrence.[8,9,15] A systematic review of relapsed SLCTs demonstrated that advanced stage was associated with poorer survival and that relapses could occur locally or at distant sites.[15] The present case therefore represents a particularly high-risk tumour because of the combination of advanced age, poor differentiation, heterologous differentiation, tumour rupture, surface involvement and extra pelvic peritoneal disease.

Surgery remains the cornerstone of treatment for SLCT. The extent of surgery depends on the patient's age, fertility considerations, tumour stage, grade and the presence of metastatic disease.[16-18] In young women with stage I disease, fertility-sparing surgery may be appropriate in selected cases. However, poorly differentiated tumours and tumours with aggressive histological features require more careful consideration. Current guidance supports comprehensive surgical management in advanced disease, with cytoreduction where feasible.[16,17] In the present case, because of the patient's postmenopausal status and extensive pelvic and peritoneal disease, radical cytoreductive surgery with hysterectomy, bilateral salpingo-oophorectomy, peritonectomy, omentectomy, bowel resection and lymph-node assessment was undertaken.

Evidence regarding chemotherapy for SLCT is limited because of the rarity of the disease. Nevertheless, platinum-based chemotherapy is generally considered for advanced-stage or high-risk SLCT.[17] BEP (bleomycin, etoposide and cisplatin) is one of the most commonly used regimens for malignant sex cord-stromal tumours. ESMO guidance recommends platinum-based chemotherapy for advanced-stage SCSTs and identifies BEP as a commonly used regimen, with carboplatin-paclitaxel as an alternative.[18] For SLCT specifically, postoperative chemotherapy should be considered in poorly differentiated tumours and tumours containing heterologous elements, even in stage I disease.[18] However, in an elderly patient, treatment selection should be individualised because BEP carries significant toxicity, particularly pulmonary toxicity associated with bleomycin. ESMO guidance specifically highlights the increased toxicity of BEP in older patients and recognises taxane-platinum therapy as an alternative.[18]

Therefore, in this 76-year-old patient, the final adjuvant treatment decision should be individualised according to performance status, renal function, pulmonary status, comorbidities, residual disease and multidisciplinary tumour-board assessment rather than applying a fixed number of BEP cycles.

CONCLUSION

This is a rare case of a poorly differentiated Sertoli-Leydig cell tumour with heterologous differentiation in a 76-year-old postmenopausal woman, presenting in an unusual manner as an obstructed umbilical hernia with acute abdomen. The absence of classical androgenic manifestations, advanced age and acute surgical presentation resulted in the ovarian malignancy being identified only during emergency laparotomy. The initial frozen-section diagnosis was a poorly differentiated malignant neoplasm with carcinosarcoma considered in the differential diagnosis, while definitive histopathology established the diagnosis of poorly differentiated SLCT with heterologous differentiation.

The case highlights several important lessons. First, an enlarging or newly symptomatic umbilical hernia in an elderly patient should not always be considered an isolated

surgical condition; an underlying intra-abdominal malignancy should be considered, particularly when associated with ascites or systemic features. Second, SLCT should remain in the differential diagnosis of ovarian malignancies even in postmenopausal women without virilisation. Third, poorly differentiated SLCT with heterologous elements may closely mimic high-grade epithelial malignancies or carcinosarcoma on frozen section, emphasising the importance of definitive histopathology and appropriate immunohistochemical evaluation.

The case contributes to the limited literature on SLCT occurring in elderly and postmenopausal women and demonstrates the importance of multidisciplinary management, careful pathological assessment and individualised treatment planning in this exceptionally rare malignancy.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

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Cite this article as: Ramesh B, Kashyap L. A case report of metastatic ovarian cancer presenting as acute abdomen with obstructed umbilical hernia. Int J Reprod Contracept Obstet Gynecol 2026;15:4634-8.