

Ovarian remnant syndrome: an unsuspected diagnosis

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ABSTRACT

Background: Ovarian remnant syndrome (ORS) is a rare condition defined by the presence of residual tissue of ovarian origin, histologically confirmed in a woman with a previous salpingo-oophorectomy, usually as a result of difficult surgery in the presence of adhesions.

Objectives: To evaluate the existing literature on ORS.

Methods: A narrative review was performed. A search for relevant articles was carried out in PubMed for the period from January 2014 to July 2024. Three original cases of ORS are also reported.

Main Outcome Measures: All available literature on the subject was analysed and articles relevant to the topic of the review were included. Additional articles were reviewed to provide an overview of the issue.

Results: A total of 10 different cases of ORS found in the literature were analysed, together with 3 original cases.

Conclusions: The presence of distorted anatomy and extensive adhesions may lead to an increased risk of residual ovarian tissue. Residual ovarian tissue may sometimes evolve into a malignant lesion. When difficult oophorectomy is suspected, the surgeon must proceed with caution to complete oophorectomy. Strict follow-up is essential to detect ORS.

What is New? This is the first narrative review including cases described in the literature and three new original cases. Our work provides a comprehensive and global view of this condition and may help in clinical practice to reduce the risk of ORS through appropriate surgical planning and possibly early diagnosis of the syndrome.

Keywords: Ovarian remnant syndrome, endometrioid ovarian carcinoma, ultrasound

Introduction

Ovarian remnant syndrome (ORS) is a rare condition defined by the presence of residual tissue of ovarian origin histologically confirmed in a woman with a previous salpingo-oophorectomy.¹ Generally, it is consequent to difficult oophorectomy in the presence of adhesions which may be subsequent to multiple surgical intervention, pelvic inflammatory disease or endometriosis, which may result in inadvertent incomplete removal of the ovarian tissue.¹ According to a previous study¹ endometriosis is the most

frequent indication for oophorectomy in women with subsequent ORS. The main presenting symptoms of this rare condition are pain and the presence of a pelvic mass but sometimes it can be an incidental finding during a routine pelvic transvaginal scan.

In the literature, data concerning the incidence of ORS are limited and for the majority based on case reports and case series, moreover malignant transformation is very rarely described.

The aim of this narrative review is to examine the current literature on this rare topic and add new data

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reporting three different cases of ORS, demonstrating how challenging the diagnosis can be and how the presentation can vary, highlighting the need to perform regular transvaginal ultrasound (TVS) also in women with previous salpingo-oophorectomy and suspect ovarian pathology also in these women. Our hypothesis is that it may be possible to find ultrasound features that may be alarming and lead the clinician to suspect this pathology, by carefully studying cases of ORS reported in the literature.

Methods

A search for relevant articles was carried out in PubMed for the period from January 2014 to July 2024. The keywords used were "ovarian remnant syndrome". Only publications written in English were included, and only studies published within the time period relevant to the research question were included in the review.

Exclusion criteria were as follows:

We excluded studies that did not fulfil the defined inclusion criteria; duplicated studies; non-peer-reviewed articles; grey literature; or reports that lacked scientific rigor.

We found 55 publications, of which 1 was excluded because the full text was not available. A total of 54 publications were identified for inclusion in the review. All titles and abstracts were carefully evaluated. In the end, 23 manuscripts were excluded because they didn't focus on the topic of the current review, and 21 others were excluded because they were animal studies.

The process followed the recommendations of the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA).² The protocol was not registered.

To provide an accurate description of the current state of the art and background of ORS, a further electronic search of the online medical database MEDLINE (accessed via PubMed) was performed to evaluate the existing literature on this condition. The titles and abstracts of the articles were carefully screened to select those relevant to our research question. We also conducted a thorough review of the bibliographies of the selected articles to identify additional publications for inclusion. All selected articles were carefully assessed for both relevance and scientific merit by three independent reviewers (I.C., A.G. and A.C.). Figure 1 shows a flow diagram of the literature search. Nine articles were selected for review (Table 1).³⁻¹¹

Case Series

Case 1 Endometrioid Ovarian Cancer: A 6-year Diagnosis

We present a case of an endometrioid ovarian carcinoma appeared in the context of a misdiagnosed ORS recognized after 6 years in the Hospital Le Scotte of the University of Siena. All ultrasonographic pictures are reported in the timeline in Figure 2.

The patient was a 65-year-old woman, with normal body mass index (BMI) (24.89) who had been in menopause for 52 years without showing any gynaecological symptoms. There was no evidence of malignancy in the patient's family and personal history. The patient had two spontaneous deliveries and had previously undergone a laparotomic appendicectomy during reproductive age.

In 2015, throughout a routine gynaecological evaluation with TVS, a multilocular cyst was detected in the left ovary,

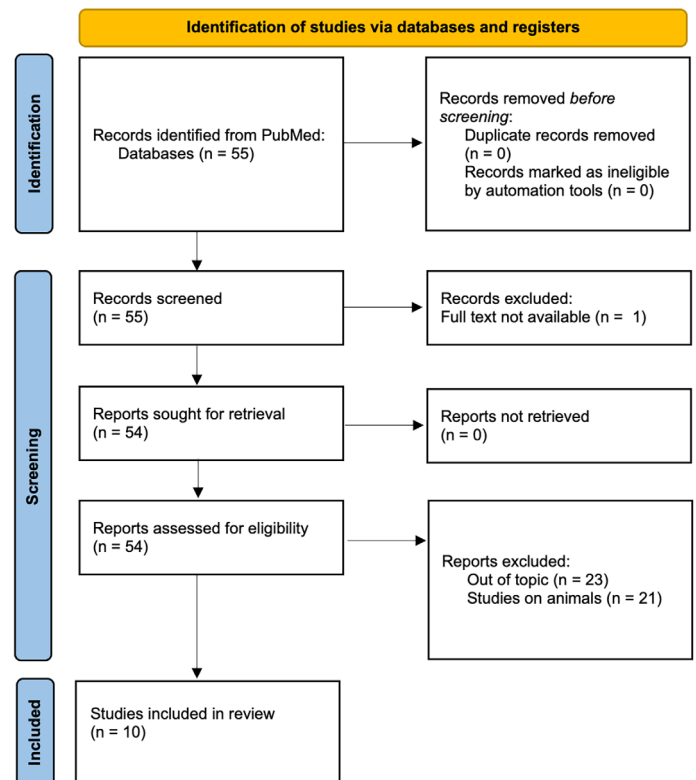


Figure 1. PRISMA 2020 flow diagram which includes searches of PubMed. Literature search diagram. A total of 55 papers filled the search string. Of these, 1 article was excluded because the full text was not available. In addition, 23 were excluded because they were out of topic and 21 were excluded because they were studies on animals. A total of 10 papers were eligible for review.²

PRISMA: Preferred Reporting Items for Systematic Reviews and Meta-Analyses.

Table 1. Main characteristics of ORS cases reported in the literature analysed in the review.

Author, journal, year of publication	Previous surgery and indication	Age at diagnosis of ORS	Case description	Treatment	Histological examination
Vilos et al. ³ , J Minim Invasive Gynecol, 2015 Case 1	Total abdominal hysterectomy and bilateral salpingo-oophorectomy Indication: extensive endometriosis Subsequent persistent right adnexal cyst that removed by laparotomy (endometriotic cyst)	50-year-old	- Right-sided pelvic pain - CT MRI revealed right adnexal cyst of 4.5x3.4x2.4 cm; severe right hydro-uretero-nephrosis - CA-125 negative	Medically treated because of complex medical and surgical history (leuprolide acetate 3.75 mg). At 12 months, the cyst, pain, and hydro-uretero-nephrosis were resolved	NA
Vilos et al. ³ , J Minim Invasive Gynecol, 2015 Case 2	Total abdominal hysterectomy followed by bilateral salpingo-oophorectomy (BSO) Indication: endometriosis	45-year-old	- Left-sided flank and pelvic pain - Ultrasound and CT identified a left adnexal cyst measuring 6x5x4 cm and moderate hydro-uretero-nephrosis	Medically treated (leuprolide acetate 3.75 mg together with oestradiol 1 mg)	NA
Gupta and Gupta ⁴ , J Midlife Health, 2016	Bilateral salpingo-oophorectomy	56-year-old	- Access to the emergency department with persistent nausea, vomiting and abdominal pain - CT revealed distended abdomen with hypoactive bowel sounds. Small, irregular soft tissue mass in proximity to site of narrowing and acute angulation of the ileal loop	Exploratory laparotomy : ascites, small bowel dilated, stricture in the proximal ileum with an adhesive band, causing near complete obstruction, small bowel mesenteric nodule	Histopathology of the mesenteric nodule associated with small bowel confirmed the presence of ovarian tissue
Chan et al. ⁵ , Cardiovasc Intervent Radiol, 2017	Total abdominal hysterectomy for adenomyosis and fibroids and bilateral salpingo-oophorectomy for endometriosis. Subsequent lysis of adhesions and attempted removal of left ovarian remnant tissue adherent to the nearby colon	44-year-old	- Chronic, constant, dull, left-sided pelvic pain - Computed tomography angiography revealed soft tissue mass in the left oophorectomy site with a volume of cc 12.5	Ovarian artery embolization	NA
Weiner and D'Andrea ⁶ , Breast J, 2018	Bilateral salpingo-oophorectomy ER + breast cancer	40-year-old 6 months after BSO	- PET revealed right adnexal cystic lesion - MRI showed 2.5x9x1.1 cm left adnexal soft tissue area and two right adnexal cystic lesions	Laparoscopy (retained ovarian tissue)	Benign ovarian tissue with focal endometriosis
Wei et al. ⁷ , Breast J, 2019	Laparoscopic-assisted vaginal hysterectomy and bilateral salpingo-oophorectomy Indication: pathogenic variant in BRCA2 in women with stage IIIA HR-positive invasive ductal breast cancer	32-year-old premenopausal woman	- Menopausal symptoms - Serum estradiol concentration 226 pg/mL - MRI showed a complex 2.5x3.1x3.8 cm right adnexal mass and a 1.4x1.3 cm	Laparoscopy	Ovarian parenchyma in the right ovarian remnant

Table 1.Continued.

Author, journal, year of publication	Previous surgery and indication	Age at diagnosis of ORS	Case description	Treatment	Histological examination
Tien et al. ⁸ Medicine (Baltimore), 2022	Total abdominal hysterectomy and bilateral salpingo-oophorectomy Indication: leiomyoma	73-year-old 30 years after BSO	- Dull lower abdominal pain for three years - No remarkable findings on TVS - On TA US cystic lesion 53.x3.3 cm in the lower abdominal region - CT of the pelvis revealed a multilocular cystic mass - CEA 3.5 ng/mL and CA-125 70.4 U/mL	Laparoscopic enterolysis and tumour excision (paraintestinal cyst with a smooth surface measuring 5×3 cm with omental adhesion to the anterior pelvic wall)	Ovarian serous cyst adenofibroma
Wills et al. ⁹ , Am Surg, 2022	Total abdominal hysterectomy with bilateral salpingo-oophorectomy Indication: unknown	68-year-old	- Abdominal pain - CT demonstrated multiple abdominal and pelvic masses, the measured 16.1×15.1×12.1	Exploratory laparotomy and mass excision (multiple masses within the small bowel mesentery)	Serous cystadenomas
Xiao and Li ¹⁰ , Asian J Surg, 2023	Prophylactic total hysterectomy and bilateral adnexectomy Indication: ovary mass and a history of breast cancer	69-year-old 2 years after BSO	- Mass at the left corner of the vaginal stump without any clinical symptoms - TVS showed a 3.7×3.3×3.9 cm septate cystic mass at the left corner of the vaginal stump, with slightly strong echo in the capsule - CA125, CA199, CEA were normal	Laparoscopic exploration and mass resection (mass bulged at the left edge of vaginal stump)	Ovarian borderline endometrioid cystic fibroma
Yao et al. ¹¹ , BMC Womens Health, 2023	Unilateral salpingo-oophorectomy Indication: umbilical cord entanglement during childbirth	47 years old 19 years before oophorectomy	- Dull lower abdominal pain for the six months preceding her presentation - Tumour mass located on the right posterior uterine wall, of 40×50 mm size - TVS showed hyperechogenic area measuring 9×10 mm in the posterior wall of the myometrium, an isoechoic area measuring 24×18 mm in the left wall of the myometrium, as well as heterogeneous hyperechogenicity measuring 48×50 mm in the anterior myometrium - CT revealed a rounded soft tissue mass approximately 46×40 mm in size within the right wall of the myometrium - CA125 181.4 U/mL, HE4 55.6 pmol/L, CA199 15.9 U/mL, CA153 10.6U/mL, CA72-4 3.5 U/mL, CEA 1.93 ng/mL, AFP 2.7 ng/mL, SCC 1.5 ng/mL	Transabdominal hysterectomy with left adnexectomy (pale-yellow mass measuring approximately 50×40×30 mm with a nodular appearance)	Clear cell carcinoma

ORS: Ovarian remnant syndrome, ER: Emergency room, HR: Hormone receptor, CT: Computed tomography, MRI: Magnetic resonance imaging, PET: Positron emission tomography, TVS: Transvaginal ultrasound, NA: Not applicable.

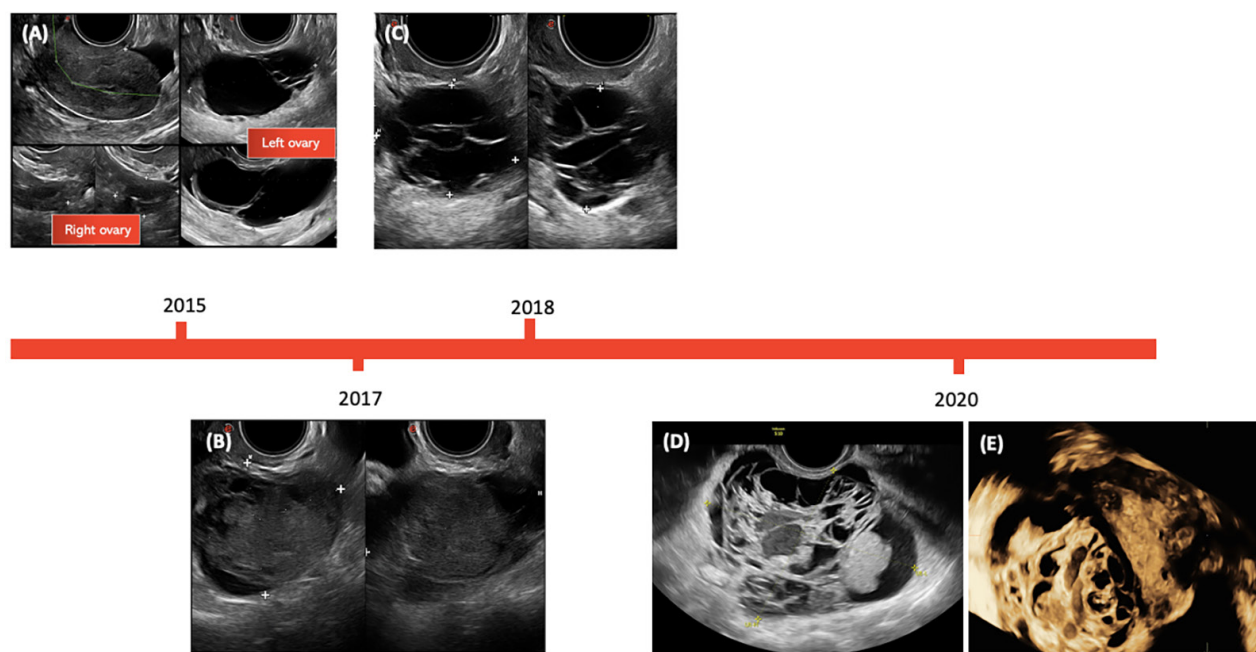


Figure 2. A-E) Ultrasonographic pelvic assessment during follow-up (case 1).

with a diameter of 74×41×56 mm, smooth internal walls, anechoic content, with colour score 1 and no crescent sign. Right ovary, uterus and endometrium appeared normal (Figure 2A).

The patient was admitted to our hospital to undergo laparoscopic bilateral salpingo-oophorectomy (BSO). The surgical report described a 70 mm cyst with fluid content that completely occupied the left adnexa and fibrotic adhesions between the colon and the adnexa retracting the uterus on the left side. No macroscopical lesions of the right adnexa and the uterus were described. The cyst was sent to histological exam, and the diagnosis was serous cystadenoma.

In 2017, the patient reported pelvic discomfort. With TVS a 71×49×62 mm solid tumour was detected located in the Douglas on the left side. The lesion was characterized by a mixed partially anechoic and partially hyperechoic echogenicity, with regular external walls, with colour score 3. The uterus and endometrium were regular. No lesions were visualized in the other side of the pelvis (Figure 2B).

The patient was admitted again to our hospital and underwent a laparotomy with the exeresis of a 60 mm retroperitoneal mass. In the surgical report it was described the presence of firm adhesions between sigmoid-colon, left ureter and left infundibulopelvic ligament which have been gently removed. The histological examination described the presence follow-up walls with hemosiderin

deposits suggestive of endometrioid cystadenoma. There was no need for further intervention and the patient was discharged with an annual follow-up visit.

In 2018, the patient underwent a follow up TVS and it was again detected a 36×29×31 mm multilocular cyst in the left adnexal region with anechoic content, smooth internal walls, not vascularized (colour score 1) (Figure 2C). Unfortunately, no tumour markers were carried out as the cyst was not investigated as ovarian cancer. We believe that with the benefit of hindsight they would be useful to guide the diagnostic process.

The referring gynaecologist decided on expectant management until, in 2020, the patient was admitted to our outpatient department for follow-up assessment. A 80×62 mm multilocular solid cyst was detected attached to the posterior uterine wall, the lesion was high vascularized at colour Doppler (colour score 2-3) (Figure 2D); its sonographic characteristics were completely different from the previous TVS.

Based on the previous history of BSO and the position of the cyst attached to the uterus, a uterine malignancy was considered in differential diagnosis (Figure 2E).

In 2021 the patient undergone a total laparotomic hysterectomy with contextual omentectomy, pelvic lymphadenectomy, rectal discoid resection and ureteral reimplantation. The invasiveness of the surgery, particularly the discoid resection, was due to the

numerous adhesions that made it impossible to dissect the lesion.

The final histologic exam diagnosed endometrioid ovarian carcinoma. The surgery was considered to be complete, and the patient did not have to undergo chemotherapy.

The patient is still under follow-up and the gynaecological assessment is negative for ovarian cancer relapses.

Case 2 Vanishing Ovarian Cyst

We present the case of a 67-year-old female patient with spontaneous menopause at the age of 52 and with a BMI of 28.1. She underwent laparotomic left ovariectomy for a dermoid cyst in 1989. Her past medical history included a previous grade IV postpartum vaginal laceration suture and a laparoscopic cholecystectomy in 2023. Prior to menopause, she reported regular, non-painful menstrual cycles. She had a long history of oestro-progestin therapy for contraception, and history of one vaginal birth.

At her annual ultrasound examination in 2023, a unilocular cyst, with anechogenic content, avascular was described on the right ovary with a maximum diameter of 1.5 cm, unchanged since 2010. The left adnexal region showed no echogenic tumefactions.

In 2024 she complained of pain in the left iliac fossa and hypogastrium with a feeling of weight in the abdomen, especially with rectal pressure.

Because of the reported symptomatology, she underwent magnetic resonance imaging, which revealed a pelvic cyst with a maximum diameter of 6.5 cm, polylobulated with modest diffuse post-contrast enhancement in the retrouterine area.

In March 2024 she underwent TVS. On TVS the right ovary appeared normal, with the known small cyst of the same size as on previous examinations. On retrouterine inspection, there was a solid multilocular cyst measuring 65x42x63 mm, which was vascularized in its solid component, with colour score 2. The presence of vascularized tissue raised the suspicion of endometrioid carcinoma or alternatively mucinous intestinal carcinoma in a possible residual ovarian syndrome. In April 2024, a further ultrasound scan was performed, and the cystic formation was no longer visible. Instead, only solid, avascular tissue resembling postmenopausal ovarian parenchyma was observed with a maximum diameter of 2 cm. Given the previous suspicion of malignancy and the postmenopausal state, the patient was referred for right

oophorectomy, peritoneal washing and exeresis of pelvic mass. In May 2024 the patient underwent laparoscopy, during which the regular uterus was visualized, with a regular right adnexus with a small cyst, while the left adnexus was absent. Posterior to the uterus, a 2 cm pelvic mass was observed, which was much smaller than on the previous ultrasound scan. Ultrasonographic pelvic assessment is reported in Figure 3. The histological examination revealed a serous cystadenoma of the right ovary, while the retrouterine formation was recognized as a fragment of ovarian parenchyma with recent haemorrhagic extravasation with associated simple cyst. In the postoperative ultrasound the adnexal fields were regular bilaterally. The patient did not require further treatment and is being followed up regularly. According to the histological report, the symptoms and the appearance of the cyst, it is reasonable to hypothesize that there has been a resumption of ovarian tissue activity despite menopause of a fragment of parenchyma remained in the Douglas, with the development of a functional formation with probable blood extravasation inside it, responsible for the internal projections visible on ultrasound control.

Case 3 the Concealed Ovary

We present the case of a 52-year-old female patient who underwent laparoscopic right adnexectomy and left salpingectomy in March 2021 for an occasionally diagnosed ovarian cyst detected on annual TVS. Her personal medical history was silent. The patient had

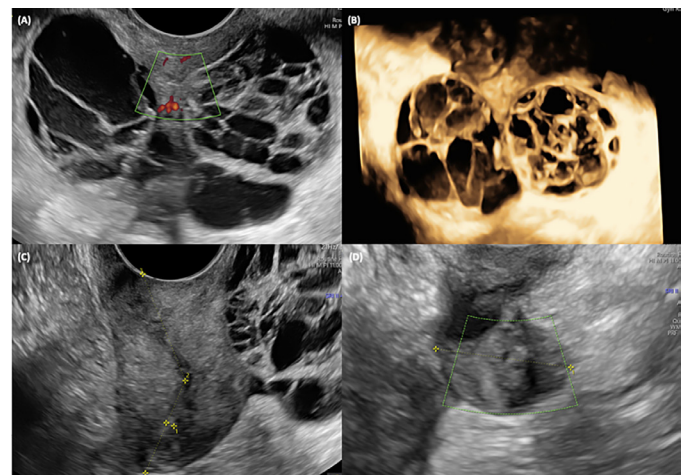


Figure 3. Ultrasonographic pelvic assessment (case 2). A solid multilocular cyst with a vascularized central solid part is observed on power Doppler (A). 3D appearance of the mass (B). The cyst was located posterior to the uterine body and cervix as clearly visible in the longitudinal scan of the uterus (C). The Pouch of Douglas was obliterated. Picture D shows the ultrasound appearance of the mass in May 2024, with the cyst no longer visible, appearing as ovarian parenchyma.

no previous surgical intervention and was completely asymptomatic.

On preoperative ultrasound, the patient presented with a unilocular cyst with anechoic content, non-vascularized on power Doppler with a maximum diameter of 8 cm. The diagnostic hypothesis suggested a serous cystadenoma. At surgery, the uterus had irregular external contours consistent with uterine fibromatosis. The left ovary appeared macroscopically normal and was attached to the ipsilateral uterosacral ligament. The right ovary appeared enlarged in volume and completely occupied by an 8 cm cyst with fluid content and regular walls, attached to the uterosacral ligament and the anterior wall of the rectum. During surgery, careful lysis of the peritoneal adhesions was performed and a right adnexectomy and left salpingectomy were performed.

Histological examination revealed an oedematous connective wall of Müllerian origin, salpinx and ovarian tissue with areas of endometriosis.

On follow-up 6 months later, a normal ovary was observed on the left and tissue compatible with an ovarian remnant on the right iliac fossa. Given the asymptomatic presentation, careful ultrasound follow-up was indicated. Ultrasonographic pelvic assessment is reported in Figure 4.

Discussion

The incidence of ORS is still unknown. The presence of distorted anatomy and extensive adhesions is associated with unfavourable surgical condition which may lead to an increased risk of ovarian tissue remnants.^{12,13}

Patients often present with chronic pelvic pain, dyspareunia, cyclic pelvic pain, dysuria and tenesmus,

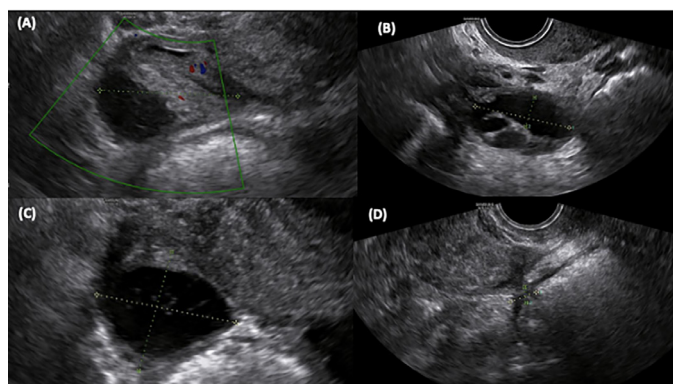


Figure 4. Ultrasound scan at 6 months (case 3). Residual ovarian tissue with follicular activity is observed (A-C). A nodule of deep endometriosis infiltrating the anterior wall of the rectum is visible in the posterior compartment (D).

caused by the growth and compression of the embedded functional ovarian tissue but they could also be asymptomatic¹² thus clinical history is fundamental in the diagnosis of ORS and a previous ovarian surgery must be recorded.

Small pieces of ovary may be functional and grow under hormonal stimulation⁸ and neovascularisation may occur.¹²

The ORS can be suspected with imaging techniques, typically a pelvic mass consistent with an ovarian remnant can be recognized in a woman who has previously undergone unilateral or bilateral oophorectomy¹⁰ but the diagnosis is only histological after surgical removal of the suspected lesion.⁸

The surgical excision of the ovarian remnant may be challenging due to the presence of adhesions, bleeding and distorted anatomy thus the procedure must be performed by an experienced surgeon and must be radical to avoid recurrences¹⁰ mainly because the residual ovarian tissue carries a risk of malignant transformation.¹⁴

In addition to anatomical distortion, a potential risk factor associated with ORS is the extension of ovarian stroma up to 1.4 cm into the infundibulopelvic ligament beyond the visible margin. Therefore, in order to prevent ORS, it is necessary to perform high ligation of the infundibulopelvic ligament, retroperitoneal dissection, and excision of all peritoneum and tissue adherent to the ovary.¹⁵

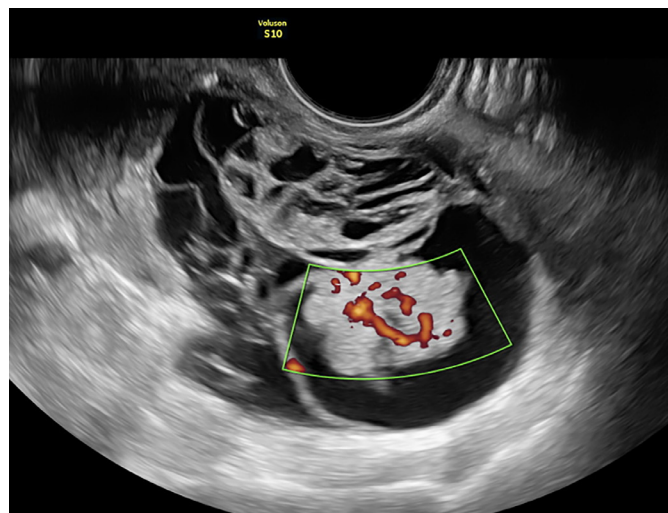


Figure 5. Ultrasonographic aspect of the endometrioid ovarian cancer in the context of ORS. It appears as a large, unilateral, multilocular-solid tumour, with anechoic cystic fluid. A large central solid component located within locules can be observed corresponding to a cockade-like sign.

ORS: Ovarian remnant syndrome.

Once a residual ovary is diagnosed, laparoscopy, laparotomy and robotic surgery can be used if surgical treatment is chosen. In a 2012 study of 223 patients with ORS, 83.9% underwent laparotomy, 8.5% laparoscopy and 7.6% robotic surgery. The laparoscopic and robotic approaches were associated with less blood loss than laparotomy and were also found to be associated with fewer postoperative complications and shorter length of stay.¹⁶

For patients who are at high risk of surgical complications or who are asymptomatic with no risk factor, conservative medical management of ORS has also been suggested. If symptoms occur, oral contraceptives, gonadotropin-releasing hormone analogues and medroxyprogesterone may be used to suppress the potential ovarian function of the remaining tissue, as well as pelvic radiation therapy. However, conservative strategies should only be reserved for cases where there is a histological diagnosis confirming ORS and excluding the risk of malignancy.⁸

One of the main causes of the presence of altered pelvic anatomy and presence of adhesions is endometriosis.

Endometriosis is a diffuse disease, characterized by the presence of endometrial tissue outside the uterine cavity, that affects about 5% of women and involves multiple pelvic organs such as the ovaries, pelvic peritoneum, pouch of Douglas, rectum, rectosigmoid, rectovaginal septum, uterosacral ligaments, vagina and bladder with different degrees of severity.¹⁷ It usually causes painful symptoms such as dysmenorrhoea, dyspareunia, dysuria, dyschezia or chronic pelvic pain, but sometimes it can be asymptomatic and may only be discovered during surgery. Nowadays, awareness of the disease has increased, and imaging techniques and knowledge have improved. TVS is the first-line imaging technique in the diagnosis of endometriosis.¹⁸ Allowing improved and quicker detection of the disease due to its wide availability, non-invasiveness and lower cost. Ultrasonographic features of lesions have been extensively described by the International Deep Endometriosis Analysis group.¹⁷ Although endometriosis is a benign disease, malignant transformation of the lesions is possible, in particular, it is associated with a higher risk of clear cell and endometrioid ovarian cancer, 3.4 times and 2.3 times respectively.¹⁹

Endometrioid carcinoma is the second most frequent ovarian carcinoma in women with a mean age at presentation of 55-58 year and up to 50% of cases develop in patients with endometriosis; it carries a 5-year survival rate of more than 70%.²⁰ Macroscopically, endometrioid ovarian carcinoma appear as a unilateral tumour

with a mean size of 150 mm.²⁰ The ultrasonographic characteristic of this kind of tumours are widely described, they generally appear as multilocular-solid tumours, with low-level echogenicity of the cyst fluid, but they also can be described as solid masses.²⁰

In case 1, with the benefit of hindsight, we can recognize in the cyst of our case most of the main characteristic features of an endometrioid ovarian carcinoma (Figure 5): a large, unilateral, multilocular-solid tumour, with anechoic cystic fluid and, if we look carefully, we can recognize a large central solid component located within locules, which may correspond to the cockade-like appearance described by Moro et al.²⁰

As mentioned above, given the patient's clinical history, the previous adnexectomy and the tight connection between the cyst and the uterine wall, in the differential diagnosis was considered a malignant pathology of the uterus, in particular a uterine sarcoma was suspected, which however has distinct ultrasonographic features. Generally, sarcomas have an irregular shape, with heterogeneous echogenicity, cystic areas and necrosis, and are highly vascularized²¹ but in complicated cases these features can be superimposable to those of an ovarian neoplasm, and they may be misinterpreted.

Unfortunately, in our case, we cannot know whether there was a diagnostic mistake in ultrasonographic evaluation and in the reading of the first histologic examination or whether the malignant transformation occurred later. Given the presence of adhesions described in the first surgery, we can speculate that the patient in the case described may have had undiagnosed endometriosis that exposed her to an increased risk not only of ORS but also of neoplastic transformation of the ovarian tissue remained in the pelvis.²²

In this case, diagnosis may be delayed because of failure in recognizing ORS which was not suspected because of the patient's previous history of bilateral oophorectomy.

Moreover, in those patients, ovarian remnant tissue may be mistakenly confused with a leiomyoma,¹¹ uterine sarcomas and adenomyomas. In most of case report in literature authors conclude, as we do, that the diagnosis is generally missed because of the patient's previous surgical history.

ORS can present with multiple histopathological diagnoses from endometrioid, clear cell but also borderline endometrioid cystic fibroma,¹⁰ mucinous adenocarcinoma,²³ ovarian serous cystadenofibroma.⁸

Surgical inattention, such as incomplete removal of ovarian tissue or morcellation in the pelvic cavity during a difficult oophorectomy, increases the risk of ORS through the dissemination of ovarian fragments into the pelvis.²⁴

In women with previous surgery, endometriosis or other conditions associated with the development of pelvic adhesions, it's fundamental for the surgeon to consider the possibility of a difficult oophorectomy and carefully proceed to a complete ovarian remove, a high ligation of the pelvic infundibulum ligaments and retroperitoneal dissection may be considered to avoid the risk of ORS.¹¹

It is preferable to remove the ovary in one block, possibly within a bag, from a larger laparoscopic port, colpotomy or mini-laparotomy, but if this is not possible and fragmentation is used, it is important to collect all the fragments and wash the pelvic cavity thoroughly. In case incomplete oophorectomy is suspected, the patient should be closely monitored to recognise the development of the syndrome in advance²⁴ and to early recognize the presence of anomalies in the adnexal area for the risk of malignant transformation.¹¹

Conclusion

ORS is a rare condition which must be suspected in case of incidental detection of pelvic mass in a woman with previous bilateral oophorectomy. The presurgical evaluation of the risk of adhesions and an accurate excision of the ovarian tissue during the initial surgery will reduce the risk of ORS.

This condition could be a completely incidental finding that is occasionally discovered during a routine ultrasound scan. If the syndrome is diagnosed, several aspects must be taken into account, from the presence of symptoms, the ultrasonographic aspect of the cyst and the patient's preference, in order to choose the correct management, from expectant management to surgery, balancing the risks and benefits of each choice. If second surgery is required, it is important that it is carried out by a team of experienced surgeons to reduce the risk of recurrences.

A possible limitation of this paper is that due to the paucity of data in the literature, it is not possible to draw conclusions. Other potential limitations of this work may be related to eventual selection bias, we attempted to include all cases described in the literature, however it is possible that some papers named with keywords not

included in our search string were not selected. However, by comparing our work with similar previous papers, we have found that the cases described are common to all studies, so we can assume that the number of erroneously omitted cases is limited.

The present paper with our case series contributes to the total number of reports and may help to provide new information on how this syndrome may manifest. We would also like to raise awareness of this possibility in a woman who has had a previous oophorectomy and is found to have a pelvic mass.

Suspect ovary even if the ovaries have been removed!

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Transparency: The manuscript is an honest, accurate, and transparent account of the study being reported; that no important aspects of the study have been omitted; and that any discrepancies from the study as planned (and, if relevant, registered) have been explained.

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