

DOI: <https://dx.doi.org/10.18203/2320-1770.ijrcog20260908>

Case Report

An unusual combination of mature cystic teratoma and ovarian carcinoid tumour: rare case report of an ovarian synchronous tumour

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Received: 14 January 2026

Revised: 12 February 2026

Accepted: 12 March 2026

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ABSTRACT

Synchronous bilateral ovarian tumours of the ovary are exceedingly rare entities characterized by the coexistence of two distinct neoplasms within the same anatomical site without histological intermixing. This case report presents a unique instance of a synchronous bilateral ovarian tumours comprising a mature cystic teratoma and an ovarian carcinoid tumour in a 46-year-old perimenopausal woman who presented with lower abdominal heaviness, a palpable mass, and right lower limb swelling. Radiological investigations revealed a large, complex right ovarian mass and a smaller left ovarian cystic lesion. A preoperative diagnosis of a neoplastic ovarian mass was made, and the patient underwent exploratory laparotomy. Intraoperative findings revealed bilateral adnexal masses; frozen section suggested benign pathology. Definitive surgery included total abdominal hysterectomy with bilateral salpingo-oophorectomy. Histopathological examination confirmed a mature cystic teratoma in the left ovary and a carcinoid tumour in the right ovary. Immunohistochemistry supported the diagnosis, showing CD56 positivity and a Ki-67 index of 15%. This case highlights the importance of considering synchronous bilateral ovarian tumours in the differential diagnosis of complex ovarian masses, especially when imaging reveals both solid and cystic components. Early recognition and accurate histopathological diagnosis are critical for guiding appropriate surgical management and ensuring optimal patient outcomes.

Keywords: Synchronous ovarian tumours, Mature cystic teratoma, Ovarian carcinoid, Neuroendocrine tumour, Bilateral ovarian masses

INTRODUCTION

A synchronous bilateral ovarian tumour is defined as the coexistence of two different tumours within the same specific anatomical site without histological intermixing.¹ They can be either benign or malignant. Although they have been reported in various organs, including stomach, oesophagus, liver, kidney, lungs, bones, and brain, they are very rarely reported from the ovaries.² Teratoma is one of the most common components of synchronous bilateral ovarian tumours reported in the ovary. Among the very few pieces of literature available, the combination usually comprises surface epithelial cell and granulosa cell

tumours.³ However, when two histologically distinct tumours arise in separate anatomical locations, they should be regarded as synchronous distinct tumours rather than a collision phenomenon. The present manuscripts report a very rare combination of mature cystic teratoma and neuroendocrine tumour of the ovary (carcinoid tumour) in a 46-year-old multiparous perimenopausal woman.

CASE REPORT

46-year-old P2L2 perimenopausal presented to gynaecological OPD with complaints of heaviness and lump in the lower abdomen for one month. It was

associated with dull, aching lower abdominal pain for 15 days. She also complained of swelling in her right lower limb for the last week. On examination, her general condition was fair and she was hemodynamically stable. She was obese (BMI 28.5 kg/m²). The right lower limb showed tense, shiny skin, was slightly erythematous, tender, and increased warmth over the swelling. The left lower limb was apparently normal. On palpation, a large abdominopelvic lump of around 22-24 weeks' gravid uterine size was felt, which was deviated towards the right side with irregular margins, firm in consistency, nontender, and mobile without any skin changes or local rise in temperature. Per speculum, the cervix was hypertrophied, with few nabothian follicles present. On a bimanual pelvic examination, the uterus was anteverted, normal size, and mobile, and right fornix fullness was felt. A large cystic mass separate from the uterus was felt, and the left fornix appeared to be free. On per rectal examination, the rectal mucosa was free. She had a B-positive blood group with an Hb of 10.2 g/dl. The patient did not exhibit any clinical features suggestive of a functional neuroendocrine tumour. There was no history of flushing, diarrhoea, bronchospasm, wheezing, or cardiac symptoms suggestive of carcinoid syndrome. All other investigations, including renal function tests and tumour markers (CCEA, CA-125, beta-HCG, and alpha-fetoprotein), were within normal limits of lysis. The pap smear was satisfactory and was negative for intraepithelial malignancy. Her ultrasound of the of the lower abdomen and pelvis revealed a complex right ovarian cyst measuring 20x18 cm with suspicion of neoplastic origin and a left ovarian cyst measuring 4x2 cm. The uterus was normal in shape and size.

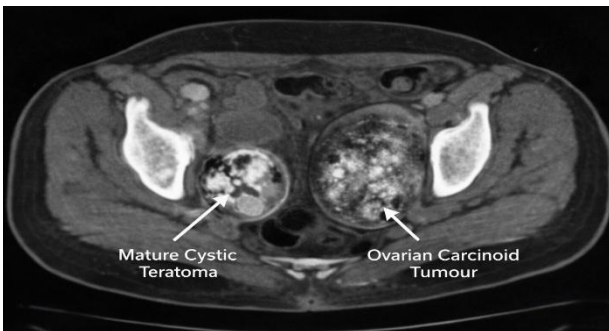


Figure 1: Contrast-enhanced CT (axial section) of the pelvis demonstrating bilateral ovarian lesions.

A colour doppler study of the right lower limb revealed the right common femoral, superficial femoral vein, popliteal vein, anterior tibial vein, and posterior tibial vein appear dilated with echogenic content within, causing partial occlusion of the lumen likely a partial thrombus. For further evaluation, CECT (W/A) was done on a large lobulated marginated complex solid cystic component lesion measuring 19.5x12.8x21.7 cm in the pelvis with superior extension into the abdomen up to the supra umbilical level as shown in figure 1. Bulky left ovary, with a focal lesion showing fat density, soft tissue attenuation,

as well as calcification within suggestive of ovarian dermoid measuring 3.8x2.6x3.1 cm. No significant pelvic or retroperitoneal lymphadenopathy was noted. The patient was managed conservatively for the right leg with anticoagulants and Sumag dressings containing magnesium sulphate and sulfacetamide. A repeat colour doppler study of the right lower limb showed dissolution of the thrombus completely. In concurrence with the senior anaesthesiologist and surgeon, the patient was subsequently taken up for exploratory laparotomy, followed by a frozen section under spinal anaesthesia. Per operative findings revealed a right ovarian mass measuring 24x21x9.5 cm, which was smooth, congested, and contained an intact capsule as shown in figure 2.



Figure 2: Intraoperative image showing large right ovarian mass.

A cut section showed a solid, creamish yellow growth with haemorrhagic areas. The left ovarian cyst measures 5.5x3.7x2.5 cm. The cut surface of the ovary was solidly cystic. Further, gross examination revealed that the cystic area was filled with cheesy pultaceous material admixed with tufts of hair. Bony and thyroid tissue were also identified. The uterus was around 10 weeks enlarged, uniformly enlarged. Multiple omental and peritoneal biopsies were taken. The intraoperative frozen section showed bilateral adnexal masses of benign aetiology; hence, it proceeded with total abdominal hysterectomy with bilateral salpingo-oophorectomy. Ascitic fluid for malignant cells revealed no definitive evidence of malignancy.

The final histopathological examination revealed a right ovarian carcinoid tumour and a left ovarian mature cystic teratoma. On immunohistochemistry, the tumour cells were diffusely positive for CD56 and focally positive for pancytokeratin. Also, the tumour cells are negative for synaptophysin, inhibin, CD99, ER, CK7, CK20, and CDX2. Ki67 labelling index was 15% in the highest labelled area. Although CD56 showed diffuse positivity supporting neuroendocrine differentiation, it is a relatively nonspecific marker. Additional confirmatory neuroendocrine markers such as chromogranin A and INSM1 would have further strengthened the diagnosis;

however, these could not be performed due to resource limitations at the time of evaluation.

DISCUSSION

Synchronous bilateral ovarian tumours are uncommon neoplasms in which elements of differing histologic origins coexist in a single mass without intermixing of tissue or cell types. Various theories and hypothesis have been postulated for the exact pathogenesis of these tumours. One such state states that the presence of the first tumour modifies the microenvironment, leading to the development of the second primary tumour or the seeding of metastatic tumours. Another theory suggests that each primary tumour results from a common stem cell. It has also been postulated that the coexistence of two different tumours in the same organ may be the result of 'chance accidental meeting'.⁴

Mostly, they present as an abdominal lump, palpable mass, lower abdominal pain, and pelvic pain. However, in some cases, it may be an accidental finding. In our case, the patient presented with heaviness and a lump in the lower abdomen, along with dull, aching pain. Ultrasound examination is the basic imaging method of choice in the initial evaluation of ovarian masses. Tumour markers are routinely used to rule out benign or malignant potential, which may include CA-125, CEA, LDH, and serum beta-HCG. Computed tomography (CT) helps in assessing the extent of disease and other morphological details. Magnetic imaging (MR) is reserved in cases where USG is equivocal and is more accurate in the diagnosis of mature cystic teratomas. The diagnosis of these tumours is based on the post-operative histopathological examination.

However, sometimes preoperative imaging can give pointers such as the presence of solid and cystic components in ovarian tumours, as shown in the multicentric study by Patterson et al.⁵ In the previously reported literature regarding ovarian synchronous bilateral ovarian tumours, the most common combinations are epithelial and germ cell tumours, followed by germ cell tumours and sexcord-stromal tumours. In the present case, a combination of mature cystic teratoma and a neuroendocrine tumour of the ovary (carcinoid tumour) was reported. Ovarian carcinoid itself is a very rare ovarian low-grade neuroendocrine tumour, accounting for about 0.1% of all ovarian neoplasms.² Thus, the coexistence of a carcinoid tumour with an ovarian teratoma in patients is

highly rare, and, our knowledge, our case may be one of the very few that have occurred in perimenopausal women.

CONCLUSION

Though synchronous bilateral ovarian tumours of the ovary are very rare, gynaecologists and pathologists should be aware of the existence of such entities. Also, the identification of different imaging characteristics within a tumour should raise a suspicion of a coexisting synchronous bilateral ovarian tumour. Identification of these tumours is essential, as it will further determine the appropriate treatment strategies based on the individual histopathological aggressiveness of each of the tumour components.

Funding: No funding sources

Conflict of interest: None declared

Ethical approval: Not required

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Cite this article as: Baghel J, Sinha M, Rawat R, Choudhary C. An unusual combination of mature cystic teratoma and ovarian carcinoid tumour: rare case report of an ovarian synchronous tumour. *Int J Reprod Contracept Obstet Gynecol* 2026;15:1401-3.