

Coexistence of benign ovarian serous cystadenoma and tuberculosis in a young woman

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ABSTRACT Genital tuberculosis involving the ovary in a non-immunocompromised individual is rare. We report a case of coexisting ovarian serous cystadenoma and tuberculosis in a 29-year-old Indian woman. Clinical examination revealed the presence of an abdominal mass suspicious for ovarian neoplasm. Histopathological evaluation revealed ovarian neoplasm and concomitant tuberculosis. To the best of our knowledge, and after an extensive search of the literature, the coexistence of benign ovarian neoplasm and tuberculosis has not been previously documented.

Keywords: benign serous cystadenoma, coexistence of benign serous cystadenoma and tuberculosis, ovarian caseous necrosis, ovarian tuberculosis, panhysterectomy

INTRODUCTION

Ovarian cystic neoplasms often present diagnostic difficulty, especially since malignant ovarian cystic neoplasms are uncommon and the determination of the locus of the malignancy requires histopathological confirmation. However, synchronous occurrence of infection and malignancy in the ovary presents an even greater diagnostic challenge. As clinical details, laboratory values and radiographic data do not usually lead to a final and definitive diagnosis, a definitive diagnosis is typically achieved histopathologically. Nevertheless, the presence of a large, multiloculated cystic ovarian mass with a central solid area on pelvic ultrasonography is often suspicious for ovarian malignancy. In our patient, histopathological study proved the mass to be a benign ovarian neoplasm in the presence of concurrent tuberculosis.

CASE REPORT

A 29-year-old Indian woman (para 2, living 2) presented to our gynaecological outpatient department with a three-month history of vague pain in the left lower quadrant of the abdomen, which was associated with a progressively increasing abdominal girth. A construction worker of low socioeconomic status, the patient had been married for 17 years and had tubal sterilisation done 12 years ago, after the delivery of her second child. She appeared pale and had a history of weight loss. Her uterus was bulky and mobile, and physical examination revealed a cystic, non-tender, mobile mass in the abdomen arising from the pelvis, with a uterine size comparable to that of a 14-week pregnant woman. Per vaginal examination revealed the presence of a mass in the left fornix, with a line of separation between the uterus and the mass. The patient had no family history of gynaecological malignancy.

Laboratory investigations and peripheral blood smear result showed a haemoglobin level of 10.4 g/dL and microcytic hypochromic anaemia with eosinophilia, respectively. The patient's erythrocyte sedimentation rate was 60 mm/hr in the first hour (baseline 20 mm/hr in the first hour). She was negative for human immunodeficiency virus (HIV). Pelvic ultrasonography revealed the presence of a multilocular, cystic, left ovarian mass, measuring 12.5 cm × 10.5 cm, with a solid component and debris. Doppler ultrasonography showed a moderate degree of vascularity with a resistive index of 0.77. As sonographic impression was that of left ovarian cystic neoplasm, confirmation via measurement of serum cancer antigen 125 (CA-125) level in the patient was required. The patient's serum CA-125 concentration was 7 µg/mL (baseline 35 µg/mL). Routine preoperative chest radiography was normal. Clinically, there were no other sites positive for tuberculous pathology, thus urine culture for tuberculosis was not performed.

The patient underwent exploratory laparotomy with a midline vertical incision. Minimal ascitic fluid was aspirated and sent for cytology and culture. A multiloculated, solid, cystic mass was seen in the patient's left ovary. Infracolic omentectomy was performed to release the adhesion between the cystic left ovarian mass and omentum, which was discovered on table. Adhesion between the urinary bladder and the left ovarian mass was also released. The right ovary was found to be minimally enlarged and cystic in consistency. Due to an intraoperative suspicion of ovarian malignancy, total abdominal hysterectomy and bilateral oophorectomy were performed, and the specimen was sent for histopathological examination.

Examination of the left ovarian mass, which measured 12 cm × 10 cm × 8 cm, revealed the presence of two cysts

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Fig. 1 Photograph shows the cut surface of the panhysterectomy specimen. The left ovary shows biloculated serous cystadenoma with cheesy caseous necrosis (arrow).

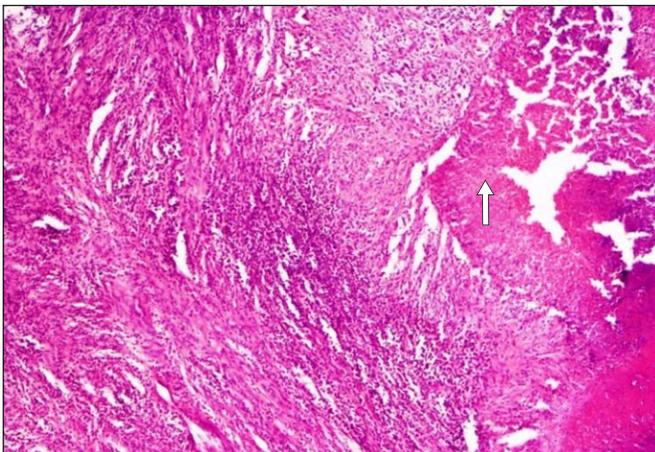


Fig. 2 Photomicrograph shows ovarian stroma with central caseous necrosis (arrow), surrounded by epithelioid and chronic inflammatory cells (Haematoxylin & eosin, $\times 100$).

with a solid area. The solid area contained cheesy, grey-white material (Fig. 1). Examination of the uterus, cervix and bilateral fallopian tubes was unremarkable. The larger cyst measured 9 cm, while the smaller cyst measured 6 cm. The right ovary, which measured 3 cm \times 2 cm \times 1 cm, appeared congested and the cut section revealed a haemorrhagic corpus luteum. Six slides were made from the cheesy, necrotic tissue of the left ovarian mass during histological examination, revealing numerous caseating, coalescing epithelioid granulomas with Langhans giant cells in the ovarian stroma (Fig. 2). There were also aggregates of lymphocytes and plasma cells. The wall of the adherent left fallopian tube showed chronic salpingitis, fused plicae and occasional epithelioid granulomas. Sections from the cystic portion of the left ovary revealed the presence of benign serous cystadenoma (Fig. 3). There was also chronic cervicitis and endosalpingitis of the right fallopian tube. Examination of the patient's right ovary and uterus was unremarkable. Ziehl-Neelsen stain of the patient's ovary was positive for acid-fast bacilli (AFB). Histological sections of the omentum

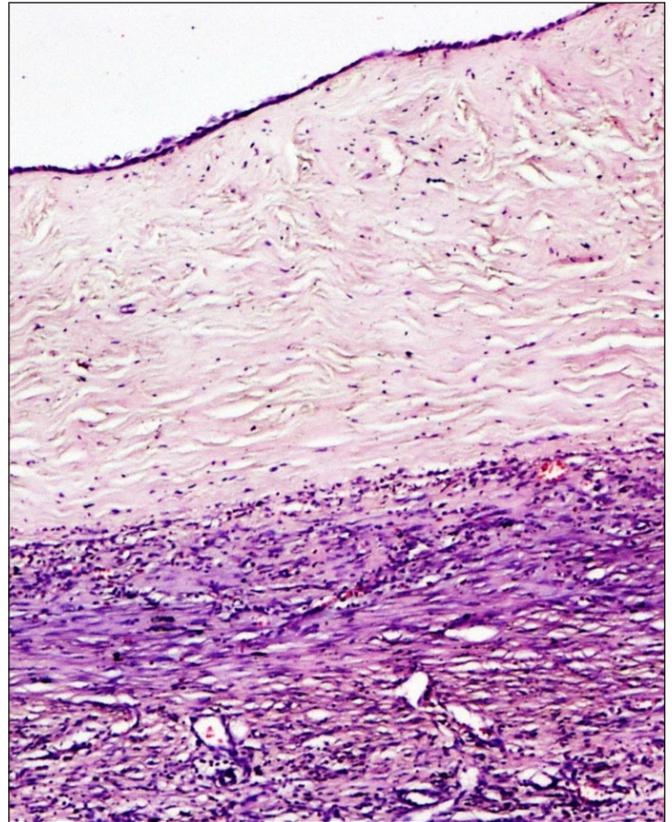


Fig. 3 Photomicrograph shows ovarian serous cystadenoma (Haematoxylin & eosin, $\times 100$).

revealed dense, chronic inflammatory infiltrate with congested blood vessels and areas of haemorrhage. Cytology of ascitic fluid showed the presence of mononuclear inflammatory cells (predominantly lymphocytes), but was negative for AFB stain and malignant cells. A diagnosis of benign serous cystadenoma of the left ovary associated with tuberculosis was rendered.

Retrospective history taking revealed a one-year history of intermittent fever occurring in the evening, associated with progressive generalised weakness and joint pain, for which the patient had repeated medical consultations. However, there was no past medical history of chronic cough with expectoration or history of contact with known cases of tuberculosis. The patient's postoperative recovery was uneventful. Diagnosed with genital tuberculosis and classified under DOTS (directly observed treatment, short-course) category I, the patient was referred to a tuberculosis treatment centre where anti-tuberculous treatment commenced.

DISCUSSION

Serous tumours of the ovary account for 20%–50% of ovarian tumours,⁽¹⁾ of which 60%–70% are benign.^(1,2) Although benign serous tumours can be both endophytic and exophytic, they are most commonly endophytic (cystadenomas), and less commonly exophytic (surface papillomas).⁽¹⁾ Grossly, serous cystadenomas consist of one or more thin-walled cysts with watery fluid, and a smooth

lining or one with soft-to-firm, polypoid excrescences that are almost entirely composed of stromata. Microscopically, these cysts are lined by either a cuboidal or columnar, nonciliated epithelium, similar to that of fallopian tubes. Psammoma bodies may be either inconspicuously present or absent.⁽²⁾

A recent report estimates the prevalence of tuberculosis in India to be at 2.186 million, with an annual incidence of 1.98 million,⁽³⁾ while Das et al⁽⁴⁾ found an approximate incidence of 18% with respect to genital tuberculosis in India. Genital tuberculosis usually occurs secondary to primary foci in the lungs, lymph nodes, bones and bowel, via haematogenous or lymphatic spread.^(4,5) Sexual transmission of genital tuberculosis has also been documented.⁽⁵⁾ Secondary disease or reactivation of tuberculosis occurs due to decreased cellular immunity. Dormant foci break down and manifest as secondary tuberculosis with the presence of caseous necrosis.⁽⁴⁾ Among the female genital organs affected by tuberculosis, involvement of the ovary has an incidence of 10%.^(2,6) Ovarian tuberculosis is usually a sequel of tubercular salpingitis.^(5,6) In the present case, involvement of the ovary was extensive, with the fallopian tube sparingly involved.

After an extensive search of the literature, we found no published account of serous cystadenoma and concomitant tuberculous infection. So far, there has only been one reported case of serous cystadenoma and suspected concomitant tuberculosis, which occurred in Poland in 1964.⁽⁷⁾ Chhabra et al⁽⁸⁾ summarised 11 cases of neoplasm with granulomas in either various organs or draining lymph nodes. In three of these cases, the absence of AFB rendered a diagnosis of neoplasm with granulomatous reaction. A stromal granulomatous reaction in the absence of tuberculosis represents a T-cell-mediated immunological response to cell surface antigens, which occurs due to a soluble, tumour-related antigen reaching the draining lymph nodes.⁽⁸⁾ However, in the present case, tuberculous bacilli with epithelioid granulomas was concomitant with ovarian neoplasm. We report unilateral serous cystadenoma involvement and ovarian tuberculosis in a young woman, which is associated with tuberculous salpingitis and the occasional presence of epithelioid granulomas in the wall of the left fallopian tube. The normal chest radiography of our patient led to a diagnosis of extrapulmonary tuberculosis, which is a rare occurrence. Ilmer et al⁽⁹⁾ has previously stated that the absence of abnormalities in the chest radiographs of patients infected with tuberculosis (up to 92%) is due to the sufficient healing ability of immunocompetent patients. Ilmer et al also highlighted HIV infection as a major risk factor for genital tuberculosis.⁽⁹⁾ Our present case is thus unique as our patient was not immunocompromised.

Preoperatively, ovarian tuberculosis was considered a remote possibility due to the absence of familial history of

tuberculosis. The patient's normal menstrual history also did not favour a diagnosis of ovarian tuberculosis. Yassaee and Farzaneh⁽¹⁰⁾ have also recorded similar observations in which the clinical impression favoured a diagnosis of ovarian malignancy, but was subsequently histopathologically proven as peritoneal tuberculosis. Additionally, serum CA-125 level has been found to be raised in the presence of pulmonary tuberculosis⁽¹¹⁾ and peritoneal tuberculosis.⁽¹²⁾ However, our patient's serum CA-125 level was within the normal limit. The diagnosis was further complicated by the intraoperative findings of ascites and adhesion to the omentum and urinary bladder, which simulated ovarian malignancy. There was also the possibility of benign serous cystadenoma and another concomitant malignant component. This case thus highlights the diagnostic difficulty of ovarian cystic neoplasm during surgery. The final diagnosis could only be confirmed on pathological examination. Histopathologically, although gross caseation is rare in tuberculous oophoritis,^(2,4) it was well evidenced in our case (Fig. 1). Under microscopic examination, the tubercular component is typically restricted to the cortex.⁽¹³⁾ However, in our case, gross evaluation of the affected ovary revealed the histological presence of cheesy, necrotic areas, and numerous caseating epithelioid granulomas with Langhans giant cells in the ovarian cortex and medulla. Positive Ziehl-Neelsen stain for AFB confirmed the presence of tuberculosis in our patient.

In conclusion, when a granulomatous lesion is associated with a neoplasm, the diagnosis becomes debatable, as concomitance of the two may be a mere coincidence. In our patient, tuberculosis had either been reactivated or reinfected in the diseased ovary. In the present case, as the size of the cystic component was much larger than the tuberculous component, revealing the presence of granulomas, we concluded that tubercular infection had occurred concomitantly with benign serous cystadenoma of the left ovary.

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