Bilateral Granulomatous Salpingitis as an Extra-pulmonary Localization of Sarcoidosis: A Case Report

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Abstract

Tubal sarcoidosis is a rare genital localization of systemic sarcoidosis. We report a case of a 52-year-old female patient with a history of ocular and mediastinal sarcoidosis, in remission for two years who presented for severe hypercalcemia. Extensive investigations showed no clear cause for the hypercalcemia ~~except~~ beyond an organic ovarian cyst. . Laparoscopy subsequently identified bilateral granulomatous salpingitis, confirming an extrapulmonary recurrence. The patient was treated with oral prednisone at 0.5 mg/kg daily for one month with a progressive taper and remained asymptomatic at one year follow-up.

Keywords: Sarcoidosis; Genital Tract; Salpingitis; Non-caseating Granuloma.

Introduction

Sarcoidosis is a chronic disease characterized by non-caseating epithelioid-cell granulomas. While it most commonly affects the lungs and mediastinal-hilar lymph nodes, 30% to 50% of patients present with extra-thoracic involvement.[1] Female genital tract localization is rarely reported.

We herein describe a case of bilateral granulomatous salpingitis discovered incidentally during management of a left ovarian cyst in a patient with known sarcoidosis and severe hypercalcemia.

Case Report

A 52-year-old post-menopausal woman with a family history of breast neoplasia and dysthyroidism, was admitted to the Department of Internal Medicine for severe hypercalcemia.

Her medical history included mediastinal and ocular sarcoidosis since 2005 which had been in stable remission for two years under treatment with corticosteroids and mycophenolate mofetil. Additionally she had been followed-up since 2018 for a persistent left ovarian cyst.

The patient presented with a one week history of polyuria, polydipsia, generalized weakness and polyarthralgia involving large joints. She reported no fever, night sweats, headaches or blurred vision. She denied respiratory symptoms, palpitations, abdominal or bone pain. She had no history of recent medication except her maintenance treatment, no history of tuberculosis and was a lifetime non-smoker.

On physical examination, she had no fever, no heart murmur, and no crackles on pulmonary auscultation. Ophthalmic examination was unremarkable with especially no uveitis or chorioretinitis.

Laboratory tests showed severe hypercalcemia with adjusted total calcium concentration of 3.52 mmol/L, and an erythrocyte sedimentation rate (ESR) of 60 mm in the first hour. No hypercalciuria was detected. Complete blood count, electrolytes and liver function tests were also normal. Based on these results, further metabolic profiling revealed suppressed parathyroid hormone (PTH) at 8 pg/mL (normal: 15-65 pg/mL) and serum 1.25 dihydroxyvitamin D at 21.28 ng/mL (normal: 30-70 ng/mL). Thyroid-stimulating hormone (TSH) test was 2.54 µUI/ml. Serum angiotensin converting enzyme (ACE) was 85.3 UI/L (normal: 30-100 UI/L). Both Mantoux and IGRA tests were negative.

Chest radiography showed bilateral hilar lymphadenopahies defining stage I on radiologic staging of sarcoidosis [Figure 1]. The thoraco-abdomino-pelvic CT scan revealed bilateral, symmetrical, non-compressive mediastino-hilar adenomegaly which were stable in number and size comparing to the last CT scans. It also showed an enlargement of the left ovarian cyst measuring 75 mm (versus 48 mm last year), uniloculated, with fluid content and a thin wall. There were no signs of solid neoplasia or haemopathy.

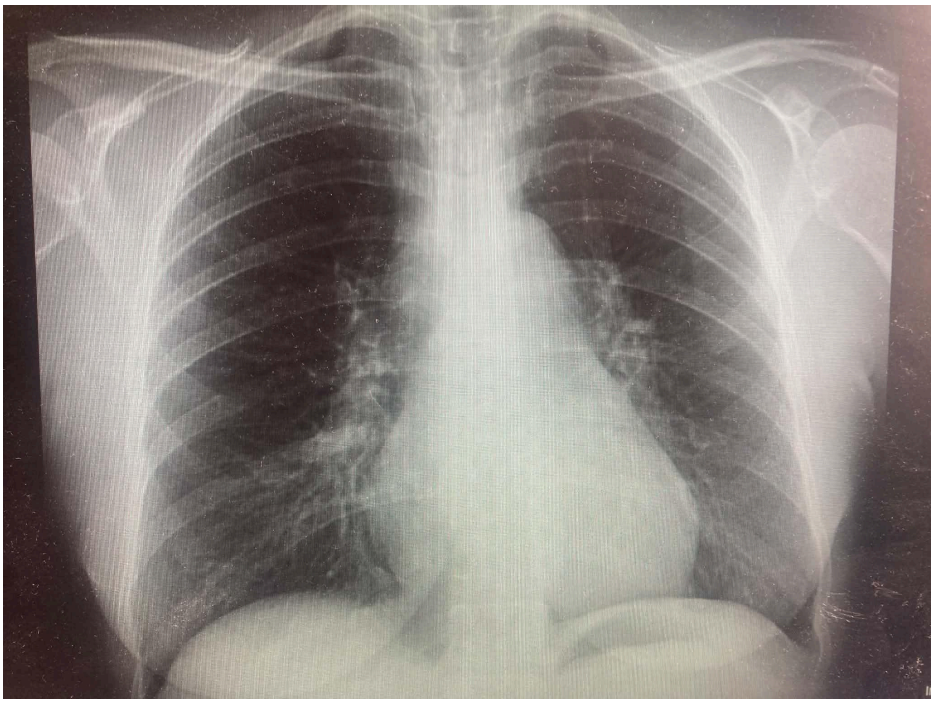

Figure 1: Chest-X-ray of the patient showing stage 1 sarcoidosis.

As no possible cause of hypercalcemia was identified including malignancy, primary hyperthyroidism, and other granulomatous diseases, the patient was treated symptomatically with hyperhydration. She was discharged after normalization of blood calcium levels and referred to her gynecologist for the management of the organic ovarian cyst.

During follow-up, the patient experienced a recurrence of hypercalcemia which correlated with a rise in serum ACE to 102 UI/L.

A laparoscopic surgical treatment for the organic ovarian cyst was performed. intraoperative observation revealed a 9 cm left paratubal cyst. Bilateral salpingo-oophorectomy was performed. Histopathological examination confirmed a left para-tubal serous cystadenoma [Figure 2]. Furthermore, the chorion of both fallopian tubes exhibited numerous non-caseating epithelioid granulomas [Figure 3, panel A, B, C]. The ovaries were free of microscopic lesions.

Based on these findings, the diagnosis of bilateral granulomatous salpingitis consistent with systemic sarcoidosis was confirmed. The patient was treated for a sarcoidosis relapse with a rare tubal localization. Management consisted of oral prednisone at 0.5 mg/kg daily for one month followed by progressive tapering. Blood calcium levels were normalized as well as ACE levels. At the one-year clinical follow-up, the patient remained asymptomatic. There were no relapses on calcium blood levels at work-up.

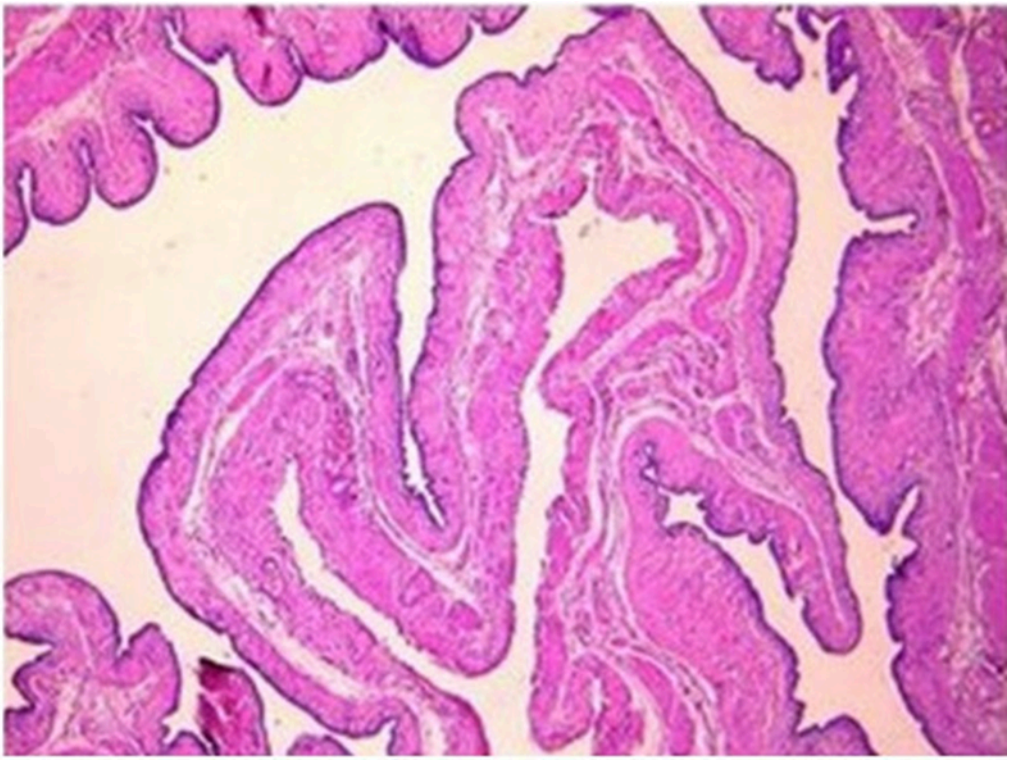

Figure 2: Microphotograph of ovarian serous cystadenoma (Hematoxylin Eosin stain x 40).

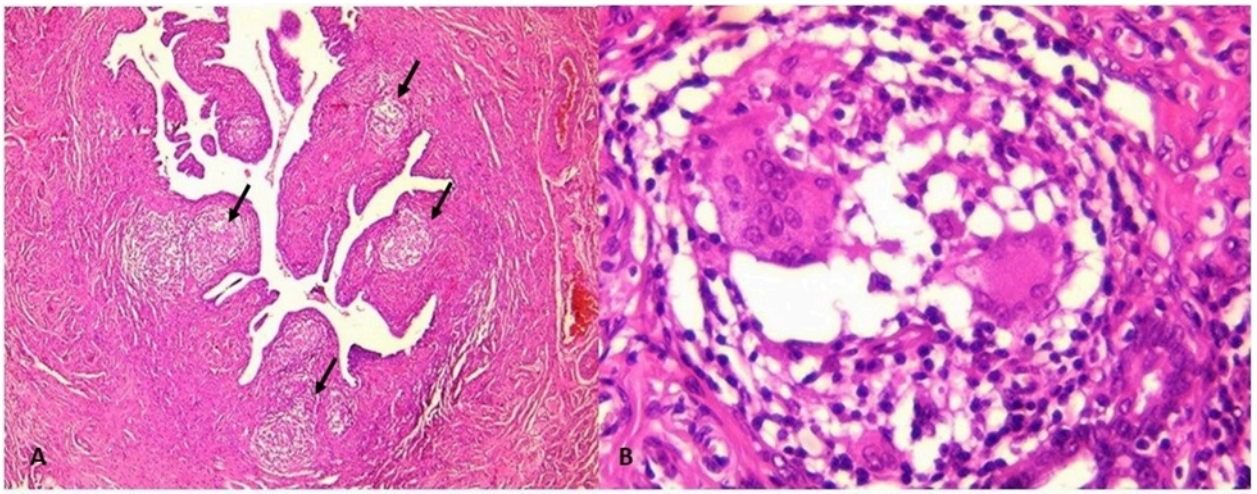

Figure 3: The tubal mucosa contains numerous small epithelioid granulomas without necrosis (A, Black arrows, Hematoxylin Eosin stain x 40) (B, Hematoxylin Eosin stain x 400).

Discussion

We reported herein a rare case of bilateral granulomatous salpingitis as an extra-pulmonary localization of sarcoidosis. The condition was diagnosed incidentally during the management of a left ovarian cyst in a patient with a known history of ocular and mediastinal sarcoidosis.

Hypercalcemia is a recognized metabolic complication of sarcoidosis occurring in roughly 11% of the patients; however severe hypercalcemia is rare, accounting for less than 5% of the cases.[2,3] In this case the

patient presented with markedly elevated serum calcium levels despite an initial lack of other clinical, radiological evidence of sarcoidosis relapse. The suppressed PTH levels correctly pointed toward an extra-parathyroid origin of the hypercalcemia.

The underlying mechanism is thought to involve high circulating concentrations of 1,25-dihydroxyvitamin D, produced by extrarenal 1α -hydroxylation of vitamin D in alveolar macrophages and active sarcoid granulomas. Unlike the renal 1α-hydroxylase, the macrophage enzyme is not influenced by PTH nor by 1,25-dihydroxyvitamin D. Instead, one of its major trophic activators is cytokine interferon gamma.[4] Lack of feedback in response to increased levels of 1,25-dihydroxyvitamin D leads to an enhanced calcium absorption in the small intestine and an excess of bone resorption leading to hypercalcemia.

Overproduction of bone resorbing cytokines and granulomatous production of parathyroid-hormone related protein (PTH-rP) may also play a role in abnormal calcium metabolism.[5]

Treatment of hypercalcemia in sarcoidosis aims to reduce intestinal calcium absorption and suppress 1,25(OH)2-D3 synthesis. Corticosteroids remain the corner stone of treatment as they are rapidly effective in normalizing calcium levels. Ketoconazole is an alternative when corticosteroids are contraindicated. It inhibits the cytochrome P450-linked enzyme systems involved in steroid synthesis including 25(OH) D3-1a-hydroxylase. Hydroxychloroquine also offers similar effects and can be considered for patients who cannot tolerate ketoconazole or who develop abnormal liver function tests. Methotrexate and azathioprine may help to control hypercalcemia by reducing the granuloma formation.[4]

Female genital tract sarcoidosis is a rare extrapulmonary sarcoidosis localization accounting for less than 1% of cases.[6] The primary differential diagnosis is infectious granulomatous disease, most notably pelvic tuberculosis.[4,7] Even if the main limitation of the case is the absence of PCR, or mycobacterial cultures on surgical specimens, the diagnosis of genital sarcoidosis was supported by the patient's established history of systemic sarcoidosis, severe hypercalcemia with no other cause, and the negativity of the Mantoux and IGRA tests.

Sarcoidosis may affect any portion of female reproductive organs. The uterus is the most frequently involved, followed by ovaries and fallopian tubes. Clinical features are variable and depend on the involved organ. Genital sarcoidosis is usually asymptomatic, but may manifest as irregular menstrual cycles, menorrhagia, metrorrhagia, infertility, ovarian enlargement, abdominal pain, obstructive uropathy, intraperitoneal mass, weight loss and ascites.[8]

Genital sarcoidosis generally carries a favorable prognosis and does not require treatment. Systemic steroids have been used to treat symptomatic uterine sarcoidosis with improvement in clinical outcome.[4]

Tubal sarcoidosis has rarely been described in the literature as only few cases have been reported. It is mostly diagnosed incidentally, based on histopathological examination of salpingectomy fragments in women treated for other gynaecological problems.[8] Table 1 summarizes the 9 cases of tubal sarcoidosis. In our patient, tubal involvement was discovered upon surgical treatment of an enlarged left ovarian cyst.

Table 1: tubal localizations of genital sarcoidosis in the literature.

Author	**year**	**age**	**discovery mode**	**Sarcoidosis diagnosis**	**Site of involvement**	**Diagnostic method**	**Associated systemic involvement involvement**
Cowdell (11)	1954	21	Autopsy	precedes	Fallopian tubes	Autopsy	disseminated granulomatous disease
Castoldi (12)	1955	44	fibroid	concomitant	Fallopian tubes	TAH/BSO	absent
Kay (13)	1956	24	Dyspareunia infertility	succeeds	Fallopian tubes	laparotomy + omentum biopsy	mediastino-pulmonary
Winslow (14)	1968	28	Cervical carcinoma in situ	precedes	Uterus, right tube and ovary	TAH/RSO	not specified

Hornoré (15)	1981	53	metrorrhagia	precedes	Uterus, tubes, ovaries	TAH/BSO	not specified
Rosenfeld (7)	1987	37	Menorrhagia	concomitant	Uterus, tubes, ovaries	TAH/BSO	absent
Boakye (16)	1997	-----	--------	precedes	Fallopian tubes	----------	pulmonary
Zurkova M (8)	2014	60	pelvic pain	concomitant	cervix, uterus, mesosalpinx, and right ovary	TAH/BSO	pulmonary
Kuno T (6)	2018	37	right lower abdominal pain	precedes	right fallopian tube	RSO	ocular and mediastino-pulmonary
Our case	2023	52	treatment for an organic ovarian cyst	precedes	Tubes	BSO	mediastinal and ocular

TAH: total abdominal hysterectomy; BSO: bilateral salpingo-oophorectomy; RSO: right salpingo-oophorectomy.

Serous cystadenoma is a common benign ovarian epithelial tumor accounting for the two-thirds of such cases.[9] These tumors typically occur in adults aged 40 to 60 years and vary significantly in size with,[10] up to 30 cm, with an average of 5–8 cm.[9] Bilateral serous cystadenoma is found in 12–23% of cases.[9]

To the best of our knowledge, only one other case of serous cystadenoma associated with tubal sarcoidosis has been reported in the literature.[8] This association seems to be fortuitous given the high frequency of serous cystadenomas and the fortuitous discovery of tubal sarcoidosis, which is often asymptomatic and only diagnosed when another gynaecological problem arises.

Conclusion

Tubal sarcoidosis is an exceptionally rare manifestation of systemic disease often diagnosed incidentally in women being treated for other gynaecological problems. Diagnosis is based on identification of noncaseating granulomas in the tubal mucosa and ruling out other conditions that can share similar histopathological findings, specifically tuberculosis. Genital tract localization in sarcoidosis should be investigated as a potential extra-pulmonary localization, particularly in patients presenting with unexplained systemic symptoms such as severe hypercalcemia.

Disclosure

The authors declared no conflicts of interest. Written consent was obtained from the patient/kin of the patient.

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