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Bilateral massive ovarian oedema – report of a case due to lymphangiitis carcinomatosa

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Summary. A 40 year-old Caucasian woman, with clinical and radiological evidence of pulmonary lymphangiitis carcinomatosa presented with weight loss, productive cough and lower abdominal pain. At operation, she was found to have massive bilateral ovarian oedema. No abdominal primary tumour was found. A transbronchial biopsy showed extensive infiltration by a poorly differentiated, focally necrotic cribriform adenocarcinoma and there was extensive lymphatic permeation by this metastatic carcinoma in the stroma of the cervix uteri, myometrium, mesosalpinx, mesoovarii, surface of the ovaries, rectosigmoid colonic wall and peritoneal fat. The cut surface of the enlarged, soft and fluctuant ovaries oozed oedema fluid readily. Histologically, there was wide separation of the stromal cells by oedema in the ovaries which contained many dilated lymphatics. Compression and collagenization of the outer cortex was noted. The pathogenesis of the massive ovarian oedema was apparently due to neoplastic obstruction of the lymphatic system.

Key words: Massive ovarian oedema – Lymphangiitis carcinomatosa

Massive ovarian oedema (MOO) due to accumulation of clear serous fluid in the ovarian stroma is a well recognised, relatively rare, entity (Kalstone et al. 1969; Massachusetts General Hospital-Case Records 1971; Kanbour et al. 1981; Roth et al. 1979; Chervanak et al. 1980; Abrams and Salazar 1981; Spinas et al. 1981; Alberda et al. 1981; Slotsky et al. 1982; Vasquez et al. 1982; Young and Scully 1984). Ovarian fibromatosis, as designated by Young and Scully (1984), with secondary accumulation of oedema fluid is closely related to MOO. Approximately 50 cases of MOO (including ovarian fibromatosis) have been reported in the literature. Young and Scully (1984) briefly commented that in their experience pelvic carcinomas with extensive pelvic and paraaortic lymph nodes involvement can occasionally give rise to MOO due to probable lymphatic permeation and obstruction draining the ovaries. A case of bilateral massive ovarian oedema due to direct and widespread lymphatic permeation and obstruction to the ovarian lymphatic drainage system by an advanced metastatic adenocarcinoma, with a probable pulmonary primary, is reported here.

Case report

A previously fit and healthy 40 year-old Caucasian female who was a life-long non-smoker presented with a 2-month history of persistent, productive cough, anorexia and weight loss. In addition, she complained of tender right ribs and lower abdominal pain. Systemic enquiries revealed no other symptoms. Clinical examination demonstrated no localised signs apart from generalised pallor. There was no obvious palpable lesions in the head, neck, breast, chest or abdominal regions.

Chest radiograph disclosed widespread nodular pattern throughout both lung fields suggestive of lymphangiitis carcinomatosa. Whole body ultrasonic scan and computerised axial tomography confirmed the lung appearance of lymphangiitis carcinomatosa with no abnormality of the liver, spleen, gallbladder, kidneys or panreas. However, the uterus was enlarged by several fibroids and the ovaries were enlarged. She had a transbronchial biopsy. A week later, she was seen by a gynaecologist who proceeded to perform a total abdominal hysterectomy with bilateral salpingo-oophorectomy. At operation, the abdominal organs were found to be normal apart from tiny nodules being present in the rectosigmoid colonic wall and the appendices epiploicae. These were biopsied. She was given Cisplatin empirically following an uneventful post-operative recovery. She deteriorated gradually and died 5 months later. A post mortem examination was not granted.

Materials and methods

The transbronchial biopsy specimen consisted of several small fragments up to 0.9 cm in maximum dimension.

The hysterectomy specimen was a 386 g uterus and cervix.

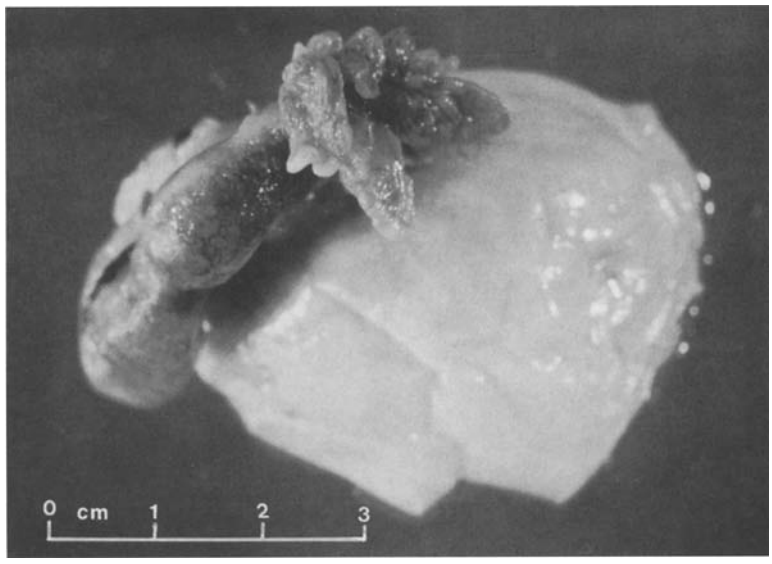


Fig. 1. Gross appearance of the external surface of the right ovary which appeared smoothly enlarged

The corpus uteri was grossly distorted by numerous intramural fibroids, ranging from 1.5 to 6.0 cm in maximum diameter. The largest one also displayed central red degeneration. The endometrial lining was not bulky. Both of the somewhat fluctuant ovaries were smoothly enlarged (Fig. 1), the right measured $6 \times 5 \times 4$ cm and the left $5 \times 4 \times 4$ cm. Both had attached attennuated fallopian tubes. The cut surface of the ovaries showed a uniform, soft, pale, gelatinous glistening surface with occasional small cysts up to 0.5 cm in diameter. Clear lymph-like fluid readily oozed out from the cut surface. There were no obvious discrete lesions in the ovaries.

The rectosigmoid colonic wall measured 0.6 cm in thickness and the fat from the appendices epiploicae contained several pale whitish tiny nodules all about 0.1 cm in diameter.

These specimens were fixed in 10% neutral buffered formalin solution. Appropriate blocks were taken, routinely processed and paraffin embedded. 4-μm thick sections were cut and stained with haematoxylin and eosin, alcian blue at pH 2.5, periodic acid Schiff's reagent with and without diastase digestion. Paraffin sections were also stained with monoclonal antibodies to cytokeratin (CK, CAM 5.2, 1 in 20 dilution, Code no: -7650, Becton Dickinson Ltd, Between Townsroad, Cowley, Oxford 0X4 3LY, England), epithelial membrane antigen (EMA, 1 in 40 dilution, Code no: -M613, Dakopatts a/s, 22 The Arcade, The Octagon, High Wycombe, Bucks HP11 2HT, England), vimentin (1 in 20 dilution, Code no: -M725, Dakopatts a/s), carcinoembryonic antigen (CEA, 1 in 20 dilution, Code no: -M30047, Oxoid Ltd, Wade Road, Basingstoke, Hants RG24 6PW, England) and human milk fat globulin-2 (HMFG-2, 1 in 40 dilution, Code no: -M21046, Oxoid Ltd) and polyclonal antibody to neurone specific enolase (NSE, 1 in 200 dilution, Code no: -A589, Dakopatts a/s) using standard routine peroxidase anti-peroxidase immunohistochemical technique. The special stains were done in conjunction with both positive and negative controls.

Results

Histologically, the transbronchial biopsy revealed extensive infiltration by a poorly differentiated adenocarcinoma displaying focal areas of necrosis and there was a cribriform pattern. Submucosal stroma showed widespread lymphatic permeation by this tumour.

The myometrium, parametrium, stroma of the cervix uteri (Fig. 2), fallopian tubes, in particular the mesosalpinx, mesoovarii showed extensive permeation in the lymphatics by metastatic cribriform, poorly differentiated, adenocarcinoma.

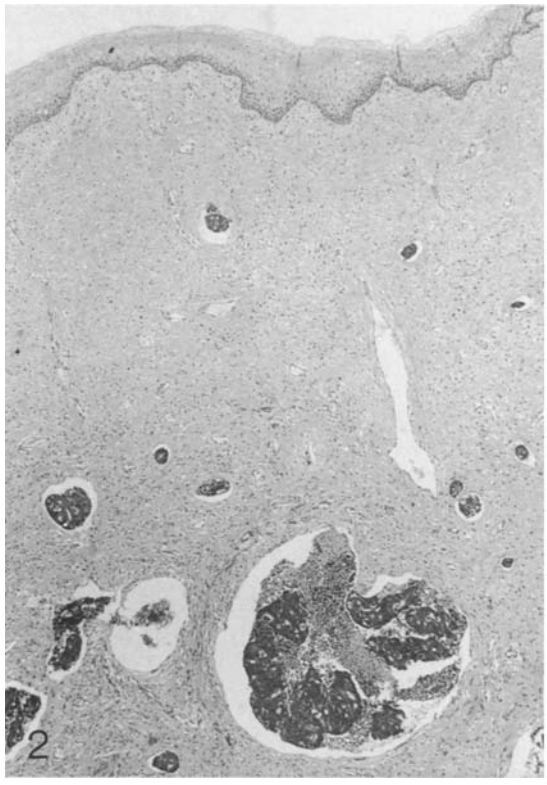
The benign nature of the intramural leiomyomas was confirmed. The endometrium appeared proliferative.

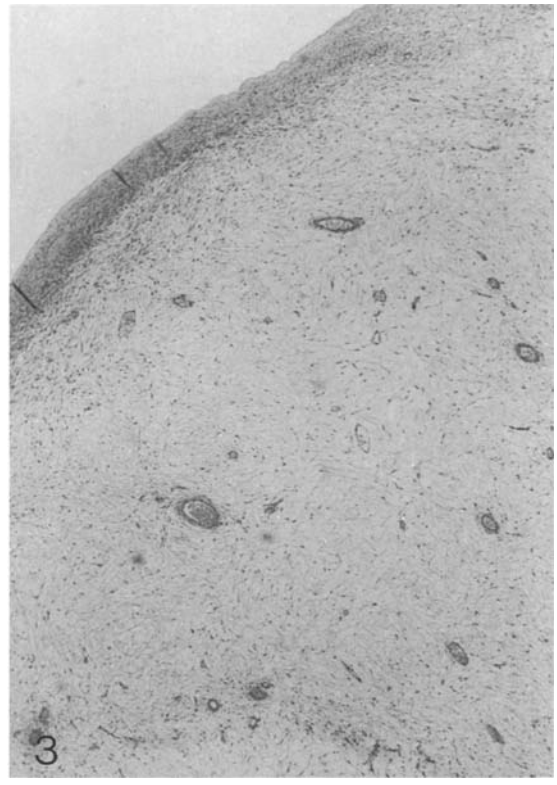
The surface of the ovaries also showed small nodules of metastatic adenocarcinomas but these were not seen in the stroma. However, the ovarian stroma was markedly oedematous with separation and clear delineation of the stromal cells by fluid (Figs. 3 and 4). The lymphatic channels were dilated. The outer cortex of the ovary was compressed and showed collagenisation. The rectosigmoid colonic muscular wall and peritoneal fat showed extensive infiltration by metastatic adenocarcinoma.

There was focal staining of PAS-diastase resistant material which was also alcianophilic, especially at the perimembranous area of the cytoplasm, in some of the carcinomatous cells. Immunohistochemistry showed the presence of strong cytokeratin and EMA cytoplasmic staining in almost all the tumour cells. In addition, focal staining of HMFG-2 and CEA was also noted in the cytoplasm of some of the cells. Vimentin and NSE staining were negative.

Discussion

There was widespread involvement of metastatic adenocarcinoma in the lymphatic channels,





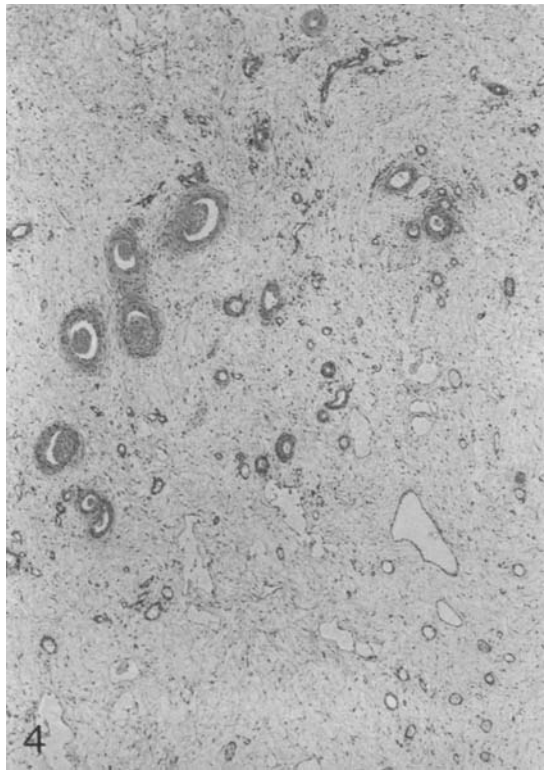


Fig. 2. Extensive lymphatics permeation by the metastatic cribriform adenocarcinoma in the cervical stroma (mag 70 ×)

Fig. 3. Note the oedematous fibrocellular stroma with a denser compact zone in the outer cortex of the ovary (mag 70 ×)

Fig. 4. The oedematous fibrocellular stroma contained dilated lymphatic channels with the smaller arteries and arterioles appearing more accentuated (mag 70 ×)

especially around the pelvic organs, in this case. Although the site of the primary tumour was not determined, in view of the findings and the absence of any primary tumour in the gastrointestinal tract, liver, pancreas, gallbladder, biliary tract and kidneys, the primary site was attributed to the lung. It is believed that this was a case of bilateral MOO due to lymphatic permeation and obstruction by lymphangiitis carcinomatosa. Young and Scully (1984) commented that pelvic carcinomas with

lymph node involvement can occasionally give rise to MOO and an acute Krunkenberg syndrome had been described by Janovski and Paramanandhan (1973) in which there was acute accumulation of oedema fluid in the fibrocelluar stroma of both ovaries.

In this case, one can lend further support to the suggestion by Kalstone et al. (1969), that MOO is due in part to lymphatic obstruction draining the ovaries. The reported cases of MOO in the literatures occurred mainly in young women. Many of these cases were partly attributed to recurrent torsion around the pedicle thereby chronically and intermitently blocking the ovarian lymphatic drainage system. In this case, however, there was clear evidence of metastatic tumour obstructing the ovarian lymphatics. It is suggested that the real clinical incidence of MOO due to lymphatic obstruction by metastatic tumour is uncertain. Lymphangiitis carcinomatosa is not uncommon in patients with widespread metastatic disease and many clinical cases of MOO may remain undetected. It is also important for the gynecologist to be aware that MOO is not always due to torsion and that, especially in older females, evidence of metastases should be suspected at the time of operation.

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