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Right Ovarian Mucinous Cystadenoma Coexisting with an Incidental Left Ovarian Haemangioma in a Postmenopausal Patient

The Editor,

Sir,

Haemangiomas were first described by Payne in 1869 (1). In the literature, their coexistence with various diseases of the female genital tract has been reported (2–5). Our case differs in that a postmenopausal woman had a mucinous cystadenoma in one ovary and an incidentally discovered cavernous haemangioma with focal positivity for progesterone receptor (PR) in the other ovary.

An 81-year old woman had right lower quadrant pain. Transabdominal sonography revealed a 10 cm right ovarian cystic and septated mass. No left adnexal lesion was reported. There was no sign of ascites. The serum cancer antigen (CA) 125 level of the patient was 144.9 IU/mL (ref 0–35), serum Ca-19-9 level was 24.9 IU/mL (ref 0–39).

Magnetic resonance images revealed a well circumscribed, slightly lobulated contoured, T1 SE hypo-, T2 SE hyperintense, multilocular cystic lesion in the right half of the pelvis (Figs. 1A–D). The left ovary measured 34 x 14 x 35 mm with slight spiculations and was isointense to myometrium on T1 SE and T2 SE weighted sequences (Fig. 1A–B). With injection of contrast material, slightly heterogeneous parenchymal contrast enhancement less than the myometrium was observed (Fig. 1A–D).

The result of the right ovary was reported as a mucinous cystadenoma on frozen section (Fig. 2).

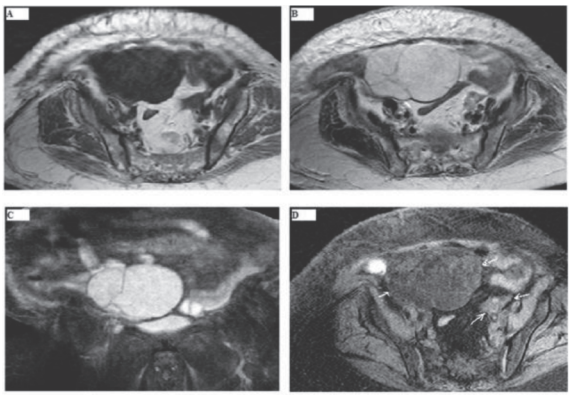

Fig. 1: Magnetic resonance imaging shows right ovarian lesion to be a multilocular cyst; (A) homogeneous hypo-signal intensity on T1-weighted images and (B) high signal intensity on T2-weighted images. Left ovary has slight spiculations isointense on T1 and T2 weighed images. (C) T2 SPAIR axial image (D) with contrast material; T1-weighted WATS CE axial images show the presence of a slightly heterogeneous enhancement in the left ovary (right lower arrow).

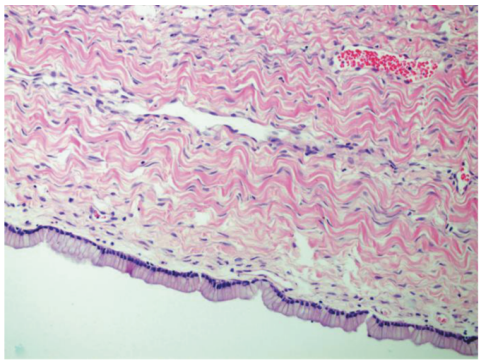

Fig. 2: Histologically, the right ovarian cyst was lined by mucinous epithelium (H&E; x 20).

On the cut surface of the left ovary (4 x 3 x 1.5 cm in size), a haemorrhagic nodular lesion, 2.5 x 1.5 cm in size, was noted. Microscopic sections revealed dilated, thin-walled vascular structures, variable in diameter, and the walls of which were comprised a single layer of endothelial cells not showing atypia (Fig. 3).

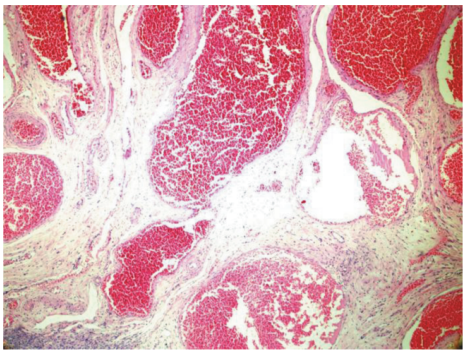

Fig. 3: Near the ovarian stroma, dilated thin walled vessels were lined by a single layer of endothelial cells, containing red blood cells in their lumen (H&E; x 4).

Based on macroscopic, morphologic and immunohistochemical findings of factor 8 (FVIII), CD 34, smooth muscle actin (SMA) expression, the case was reported as ovarian cavernous haemangioma. Oestrogen receptor (ER) was negative; however, some endothelial cells were positive for PR (Fig. 4).

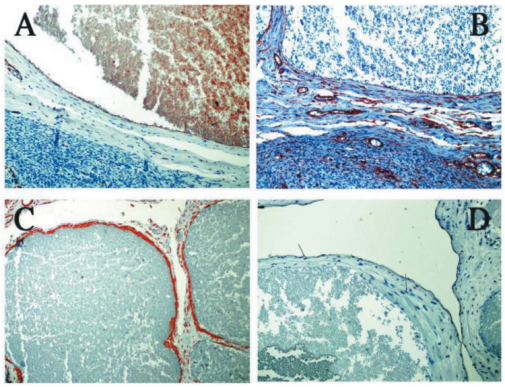

Fig. 4: Immunohistochemical findings; (A): strong positivity for factor 8 and (B): CD 34 in cells lining the cavernous vessels; (C): immunoreactivity for smooth muscle action in the walls of the vessels; (D): focal positivity for progesterone receptor in the endothelial cells (arrow; x 20).

Ovarian tumour coexisting with an incidental haemangioma has been previously reported (2–5). A review of the literature reveals that the present case was different in terms of capillary haemangioma in a postmenopausal patient with positive PR receptor (Table).

The ovarian hilus had a histological appearance that is often confused with haemangiomas due to its rich vascular structure. A mass composed of large vascular channels, nodular formation and minimal amounts of stroma might be helpful to distinguish haemangiomas from dilated, hilar vascular structures (6). In the present case, the mass was composed of dilated blood vessels with little stroma and this helped to establish the diagnosis of haemangioma.

The pathogenesis of stromal luteinization remains complicated and it is still controversial whether luteinized stromal cells promote the development of haemangiomas (7). Some authors have studied immunohistochemical expression of ER and PR, and they have found different results (3, 7). In the present case, luteinization was not observed. Moreover, endothelial cells were negative for ER, but some endothelial cells were positive for PR.

Radiological appearance for the left ovary was inconclusive; the presence of an enlarged ovary (8) and visualization of contrast uptake on preoperative magnetic resonance imaging in a postmenopausal woman supported rich vascular structure. In postmenopausal women, enlarged ovaries with or without contralateral lesions should be evaluated cautiously.

Keywords: Magnetic resonance imaging, MRI, mucinous cystadenoma, ovary hemangioma, postmenopausal women, progesterone receptor

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Table: Clinicopathologic features of patients with ovarian haemangioma and coexistent lesions reported in the literature

Author	Age (years)	Symptom	Size (cm)	Type	Luteinization ER/PR	Coexisting Lesion
Jurkovic *et al,* (3) 1999	32	Asymptomatic	NA	Capillary	No ER-PR-	Mucinous cystadenoma
Akbulut *et al,* (2) 2008	65	Irregular vaginal bleeding and pelvic pain	0.5	Capillary	No ER-PR-	Serous papillary carcinoma
Comunoglu *et al,* (4) 2010	81	Hypertension and hypornatremia	3.5	Cavernous	NA	Mature cystic teratoma
Current case, 2013	81	Lower right quadrant pain	2.5	Cavernous	No ER-PR focal+	Mucinous cystadenoma

NA: Not available ER/PR: oestrogen/progesterone positivity

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Can Chronic Consumption of Noni (*Morinda citrifolia*) Juice Lead to Changes in the Coagulation Profile, Deranged Liver Function and Excessive Intraoperative Haemorrhage?

The Editor,

Sir,

Excessive haemorrhage in orthopaedic surgery can be due to multiple causes. One of the rarer causes is the chronic effect of certain drugs (1). Ascertaining preoperative history and stoppage of use of platelet inhibitors, thrombolytics or anticoagulants is imperative (2). Occurrence of excessive bleeding, despite the application of a tourniquet, can be mystifying.

A 68-year old hypertensive female was posted for removal of intra-medullary nail with a discharging sinus from the left tibia, under general anaesthesia. The investigations were normal except for elevated lactate dehydrogenase (LDH) of 401 U/L, prothrombin time (PT) of 16.7 seconds and international normalized ratio (INR) of 1.6.

In spite of tourniquet inflated to 300 mmHg intraoperatively, there was uncontrolled, continuous and profuse bleeding, amounting to nearly 1200 mL. The intra-medullary nail removal had to be abandoned. However, during debridement of the lower tibia, the patient continued to ooze, with the total blood loss estimated to be 1500–1600 mL. The bleeding was assessed as Grade 5, according to Fromme-Boezaart surgical field grading (3). The surgery lasted two hours with the tourniquet time of 77 minutes. Postoperatively, patient informed that she frequently consumed "noni" juice for the last six to seven years. When on noni, she also experienced vaginal bleeding which mimicked menstrual periods and stopped when noni was discontinued. Fifteen days after surgery and discontinuing consumption of noni, repeat investigations were within normal limits: PT 10.5 seconds, INR 1 and LDH 151 U/L.

People use herbal/animal products for various purposes. The commonest examples are ergot alkaloids (4), belladonna (5) and ginseng (6). Evidence suggests excessive bleeding in patients consuming ginkgo biloba (7, 8) due to ginsenosides, which inhibit platelet aggregation by inhibiting cyclooxygenase/thromboxane A2 synthase, needed for platelet function (7).

All parts of noni (*Morinda citrifolia*) are used to make "bush" medicine (9, 10). Noni juice contains multiple alkaloids (10) with anti-inflammatory activity mediated *via* inhibition of cyclooxygenase/lipooxygenase which is inherently associated with the process of coagulation. These are also suppressed by ginsenosides (7, 8) and non-steroidal anti-inflammatory drugs [NSAIDs] (11). In addition, chronic consumption of noni juice does cause hepatotoxicity [elevated enzymes, deranged coagulation profile to fulminant hepatic failure requiring liver transplant] (12, 13).

In our patient with chronic consumption of noni juice, occurrence of vaginal bleeding, significantly prolonged PT/INR and high levels of LDH suggest alterations in the coagulation profile, probably *via* hepatic enzymatic synthetic mechanisms and derangement in the liver function. The return of these findings to normalcy on stopping noni further strengthens our viewpoint.

We conclude that chronic consumption of noni may actually prolong PT/INR, elevate hepatic enzymes and such a patient may have excessive intraoperative haemorrhage. Eliciting a history of consumption of herbal products like noni, assessment of coagulation profile and liver enzymes, stoppage of noni juice and waiting until the PT/INR/liver functions become normal is recommended.

Keywords: Chronic consumption, elevated liver enzyme levels, excessive intraoperative haemorrhage, Morinda *citrifolia*, noni, noni juice, orthopaedic surgery

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