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Incidental Finding of an Oestrogen-Secreting Calcified Stromal Luteoma in a Post-Menopausal with Vaginal Bleeding: An Unusual Case Report

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Abstract

Luteomas are a rare non-neoplastic tumour, which usually occur during pregnancy under the influence of human Chorionic Gonadotropin (hCG) [1]. Most cases resolve completely postpartum [2]. Patients are usually asymptomatic with the ovarian enlargement being incidentally discovered during examination, imaging or surgery. Androgen secretion by these tumours cause symptoms of hyperandrogenism in the female and virilization of female foetuses during pregnancy. Progesterone and oestrogen secretion has also been described and post-menopausal vaginal bleeding is the most frequent clinical manifestation3. We present a 75-year-old female with a left ovarian mass, discovered incidentally at the time of physical examination for post-menopausal vaginal bleeding.

Case presentation

Clinical Presentation

A 75-year-old multiparous postmenopausal woman, who presented initially with a 2-month history of vaginal bleeding and pelvic organ prolapse. Examination revealed a tender left iliac fossa mass and cervix protruding through the introitus. Her past medical history was right breast cancer with lumpectomy, the previous year before her new symptoms.

Investigations and treatment

A transabdominal and transvaginal ultrasound showed a thickened and cystic uterine endometrium. There were limited views of the adnexa due to much overlying bowel gas, hence, the right ovary was seen with a transabdominal approach only, the left ovary was not identified. Due to the findings, endometrial biopsy was performed and the histology was consistent with fragments of benign endometrial polyp and atrophic tissues. There was no evidence of atypical hyperplasia or malignancy at this time.

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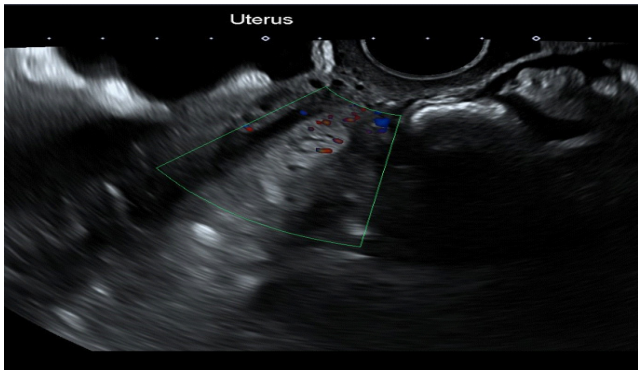

Figure 1: longitudinal view TV ultrasound showing a thickened and cystic endometrium.

CT Abdomen and Pelvis with Contrast done to further elucidate the left iliac fossa mass reported a calcified enhancing lesion that appeared to be vascular in nature measuring 38 mm AP x 41 mm transverse in the left adnexa likely to represent a left uterine artery aneurysm/arteriovenous malformation. A calcified left tubo-adnexal lesion was in the differential though was thought to be less likely. Prominent endometrial cavity measuring 6 mm was also noted. There were no enlarged pelvic or para-aortic lymph nodes.

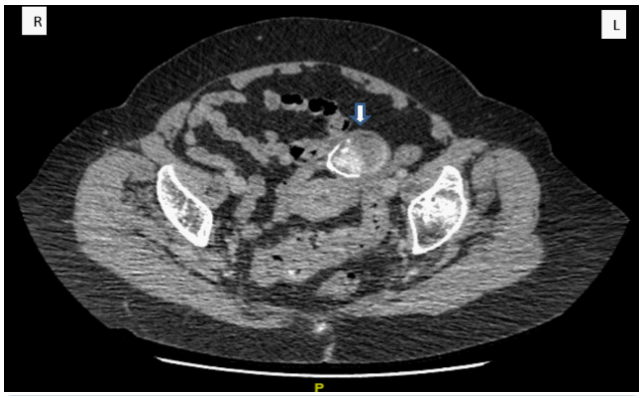

Figure 2: Axial view CT Pelvis with contrast showing left calcified adnexal mass.

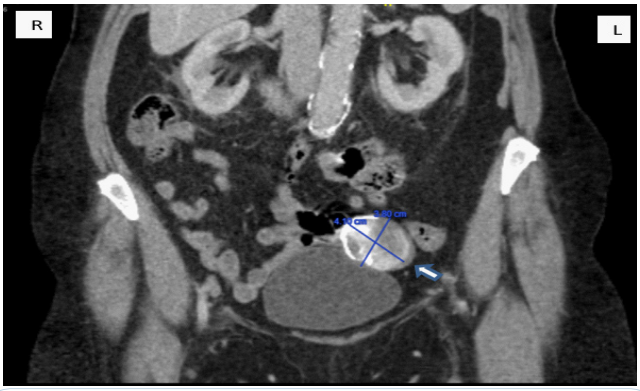

Figure 3: Coronal view CT Abdomen and Pelvis with contrast showing left calcified adnexal mass.

Due to the uncertainty of the CT findings, an MRI of Pelvis was performed which showed the mass had a well-defined low signal rim with an apparent area of high signal on T1 and T1FS posteriorly within it suggesting either haemorrhage or proteinaceous fluid and a tiny area of fat anteriorly; otherwise, it was a mixture of low and intermediate signal on T2 and fairly bland intermediate signal throughout on T1. There was no enhancement post gadolinium. It was concluded as stable left adnexal mass which had benign features but diagnosis was uncertain.

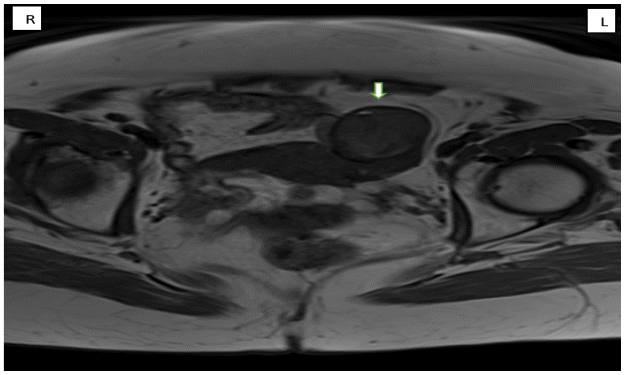

Figure 4: Non-contrast MRI Pelvis: Axial T1.

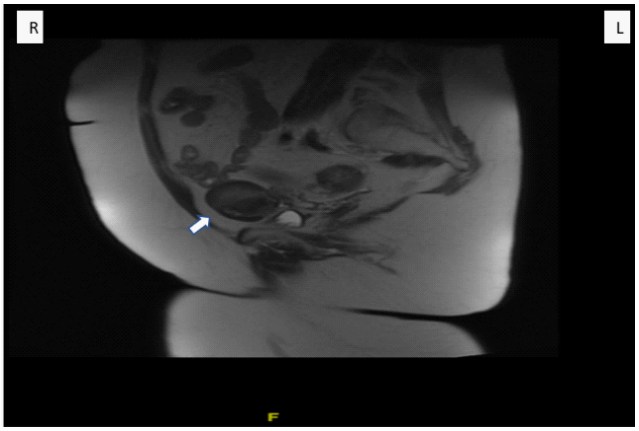

Figure 5: Non-contrast MRI Abdomen and Pelvis: Sagittal T2.

Post contrast T1FS acquisition was obtained; No enhancement was demonstrated within the lesion which confirms probable benign aetiology.

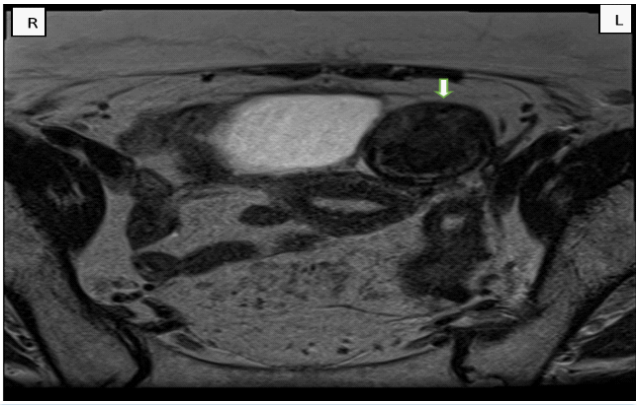

Figure 6: Oblique Axial T2 along ovarian axis.

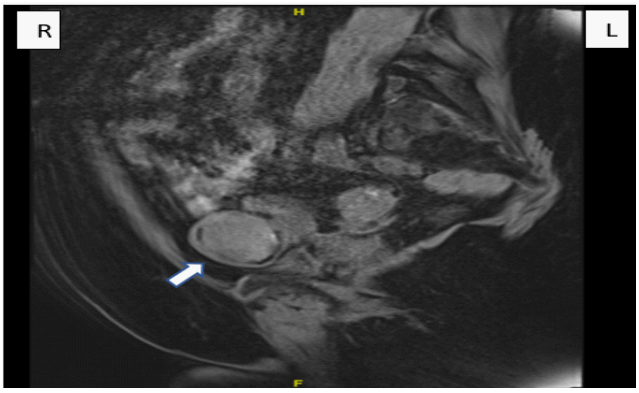

Figure 7: Contrast enhanced (post gadolinium) sagittal T1 FS demonstrated no enhancement.

Blood levels of CEA, CA 125, inhibin A and B were all normal. Despite the benign histology reports from initial endometrial biopsy and the imaging findings, her symptoms of vaginal bleeding continued and the pelvic organ prolapse worsened leading to examination findings of procidentia with fresh blood over the cervix. A decision was then made to carry out a vaginal hysterectomy and left oophorectomy. Histology of left ovary; macroscopically, measured 48 x 40 x 35 mm, cyst appeared intact, external surface appeared smooth. Calcified area of ovary measuring 35 x 35 x 23mm (not sampled), specimen calcified on sectioning contained tan coloured creamy material. Microscopic findings of small nodular proliferation of polygonal cells at the hilum. These cells showed vesicular nuclei and moderate eosinophilic cytoplasm. The calcified cystic lesion was lined by steroid/luteal cells. In addition, separate nodules of luteal cells were also seen. The appearances were in keeping with a calcified luteoma noted to have secreted oestrogen.

On histology of the uterus; it measured (three dimensions) 85 x 45 x 30 mm, endometrial polyp was identified measuring 7 x 6 mm. Tumour type was Endometrioid. FIGO grade 1 with Myometrial invasion of <50%. Tumour-free distance to serosa (mm): 9. This case, to our knowledge, is the first case report of an oestrogen secreting luteoma which likely have predisposed the patient to the breast cancer and endometrioid-type endometrial cancer Grade 1 Stage 1a.

Subsequent decision following these histological findings was for a right salpingo-oophorectomy which was subsequently done and revealed normal histological limits with para-tubal simple serous cysts. There was no evidence of malignancy.

Differential diagnosis

Granulosa stromal tumours with sex cord elements

- Adult type granulosa cell tumours
- Juvenile type granulosa cell tumours
- Sertoli-Leydig granulosa cell tumours
- Sex cord-stromal tumours of mixed or unclassified cell types
- gynandroblastoma
- sex cord tumour with annular tubules (SCTAT)
- unclassified

Pure stromal tumours

- Thecoma
- Fibroma
- Fibrosarcoma [4]

Sertoli stromal cell tumours

- Sertoli–Leydig cell tumours
- Sertoli cell tumour
- Stromal–Leydig cell tumour [5]

Other ovarian stromal tumours

- Sclerosing stromal tumour
- Signet-ring stromal
- Microcystic stromal tumour
- Ovarian myxoma
- Stromal–Leydig cell tumour

Steroid cell tumours

- Stromal luteoma
- Leydig cell tumour – Hilus Cell tumour, Leydig cell tumour (nonhilar type)
- Steroid cell tumour [4]

Discussion

Luteomas are a rare non-neoplastic tumour, which usually occur during pregnancy [1]. Aetiology of luteoma is unclear: It is thought that it arises from the proliferation of luteinised cell under the influence of β-hCG [6]. Most cases resolve completely postpartum [2]. Patients are usually asymptomatic with the ovarian enlargement being incidentally discovered during examination, imaging or surgery. Androgen production by these tumours cause symptoms of hyperandrogenism in the female and virilization of female foetuses during pregnancy. Progesterone and oestrogen production has also been described and post-menopausal vaginal bleeding is the most frequent clinical manifestation [3]. Radiologic recognition is important so that unnecessary oophorectomy, with concomitant risk to both the patient and the foetus, is avoided. As well as, prompt surgical intervention for persistent tumours, particularly in post-menopausal women, to reduce the risk of excessive oestrogen exposure and resultant endometrial, breast and ovarian carcinomas which was the case in this index patient.

Histologically, luteomas may vary, and can measure up to over 20 cm in diameter. Macroscopically, features are soft, reddish tan, fleshy, circumscribed areas with frequent foci of haemorrhage [6]. Microscopic examination shows the presence of sharply circumscribed rounded masses of polygonal cells arranged in sheets, cords, or small clusters or form containing pale fluid or colloid-like material [7]. The cells possess eosinophilic cytoplasm, large round nuclei, may be pleomorphic, and have prominent nucleoli. Common to have Mitotic figures (up to 7 per 10 hpf). The cytoplasm has less lipid and typically immunoreactive for inhibin. The cells form masses divided by the reticulin fibres into clusters, compared with the histologically similar theca cell tumour where reticulin borders each cell. When degenerate postpartum, the lipid content of the cells increases. The luteoma is reduced to shrunken nests of degenerating cells mixed with lymphocytes and fibrosis [8]. The histology characteristics of pregnancy luteoma agree with the findings in our case.

Radiological features of luteoma are difficult to differentiate from other ovarian tumours because of its solid nature. Ultrasound scan luteoma appears as a solid mass which can be single, or multinodular. It may be unilateral or bilateral with a cystic appearance due to the presence of haemorrhagic foci [9].

The MR imaging findings of pregnancy luteoma have been reported in the literature; there was a case of bilateral multilobulated cystic ovarian masses with thick septa that were incidentally identified in an asymptomatic woman at 32 weeks of gestation [10]. Another case of bilateral multinodular masses that were distributed peripherally in both ovaries. These masses were characterized as intermediate high signal intensity on

the T1-weighted images and low signal intensity on T2-weighted images which were presumably contributed to by the colloid material in the cells [11]. This is consistent with our findings on MRI. The Gd-DTPA enhanced T1-weighted images also showed avid enhancement of the masses that indicated their solid nature and hypervascularity [11], which was not the case with our report.

Learning Points

- The radiologist should be familiar with the imaging features of ovarian tumours.
- Post-menopausal vaginal bleeding and pelvic adnexal mass should prompt investigation for hormone secreting tumours such as luteomas.
- Radiologic recognition is important as unnecessary oophorectomy can be avoided in pregnancy and young females.
- Surgical removal of persistent lesions, especially in postmenopausal women, can reduce the risk of oestrogen secreting tumours and evade endometrial, breast or ovarian malignancies.

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