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CASE REPORT

Endometrial Vascular Dystrophy and Isthmocele: Hysteroscopic Visual Enigma

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Introduction

Thirty-three-year-old P1L1 with previous cesarean section had heavy menstrual bleeding and intermenstrual spotting since 1 year. Her cesarean section was done 5 years back due to non-progresss of labor. Ultrasound depicted an anechoic triangular defect in the myometrium. MRI showed gaping cesarean scar with a markedly thinned out overlying myometrium and a cystic lesion of 18.4 × 18.0 × 10.0 mm. She was diagnosed to have isthmocele along with a 3 cm posterior wall adenomyoma. Patient expressed her desire for future fertility hence a laparoscopic isthmocele resection and repair along with endometriotic cystectomy was planned for her. Informed consent was taken from the patient. Ethical clereance was taken from institutional committee. On hysteroscopy (Fig. 1), serpiginous glands were seen all over the endometrial cavity as seen in endometrial vascular dystrophy. A pouch like defect in left lateral wall at isthmus and previous scar was seen and bilateral ostia were normal.

On laparoscopy (Fig. 2), omentum was adherent to anterior abdominal wall and over previous scar. Uterus was enlarged up to 8 weeks, adenomyotic with multiple myomas with isthmocele defect; bilateral tubes were healthy. Right ovary had with multiple endometriotic cysts, 5 × 5 cm and left ovary had single endometriotic cyst, 3 × 3 cm. On chromopertubation, bilateral free spill was seen. Bladder was densely adherent to previous scar, it was dissected off by sharp dissection. Scar excision was done and edges were freshened. Uterus scar was repaired with V-Loc 1–0 in 2 layers. Three fibroids (type VII) 1 × 1.5 cm near right cornu, 1 × 2 cm on anterior wall, 1 × 1 in posterior wall and adenomyoma (type VI), 3 × 3 cm in left postero-lateral wall were seen. Vertical incision was given in posterior wall after injecting vasopressin. Myomectomy and adenomyomectomy were done along with bilateral endometriotic cystectomy. Endometriotic spots in pouch of Douglas and posterior wall of uterus were fulgurated. Myometrial defect was repaired with V-Loc. Bladder integrity was checked and foley's catheter was inserted in uterus and kept for twenty-four hours.

Postop period was uneventful and patient was discharged on day two of surgery. Histopathology revealed secretory endometrium and endometriotic cyst along with adenomyosis.

Jacques E. Hamou first described endometrial vascular dystrophy in 1991 [1] as a rare hysteroscopic finding. He discovered two types of vascular dystrophy, first was homogenously spread spiral like vessels and other was meshed or branching capillaries breaking up into the subepithelial plexus just underneath the basal endometrial layer. The exact pathogenesis of vascular dystrophy is still not clear and is more often reported in women scheduled to undergo hysteroscopy during the luteal phase of the menstrual cycle and in patients receiving progestogen therapy. Although the differential diagnosis of this variant includes Rendu–Osler–Weber disease [2] in which telangiectasias and arteriovenous malformations of the skin mucosa and viscera are seen, no such cases have been reported in the literature. Paoletti et al. have reported two cases of endometrial vascular dystrophy, first in an infertile women with endometrioma where on hysteroscopy the entire endometrial

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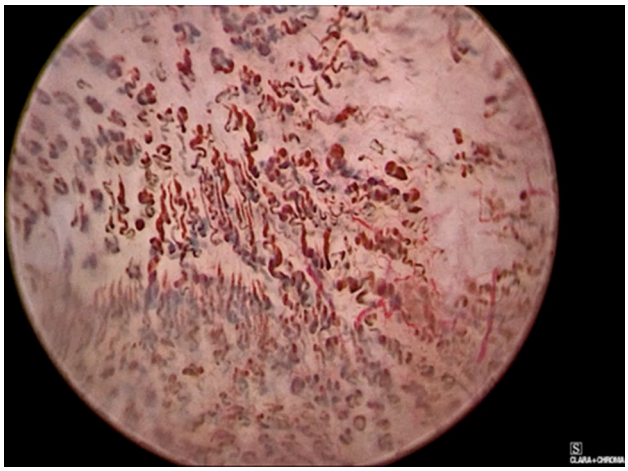

Fig. 1 Hysteroscopic image of the tortuous serpiginous endometrial glands at the fundus suggestive of vascular dystrophy

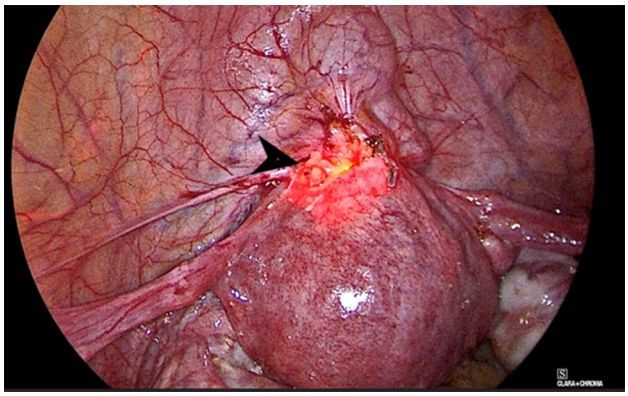

Fig. 2 Laparoscopic view with hysteroscopic transillumination amplifying the borders of isthmocele (marked with black arrowhead). Rest of the uterus was adenomyotic

lining seemed to be covered by reddish tortuous vessels arranged in a parallel pattern. Second case was a 33-year-old woman referred for management of spotting and polymenorrhea. Transvaginal ultrasonography demonstrated a focal thickened endometrium suggestive of endometrial polyps and hysteroscopy depicted the presence of a 1-cm endometrial polyp, and several reddish tortuous vessels arranged in a parallel pattern were detected in the fundal area. Multiple endometrial biopsy specimens in both cases revealed secretory endometrium with stromal decidualization. He reported the spontaneous disappearance of these findings in the following cycle, thus advising avoidance of unnecessary intervention in these cases.

But on the other hand, Sopelana et al. reported eight cases of similar diagnosis in their retrospective review of 7658 cases over a span of 6 years and concluded that endometrial vascular dystrophy does not comprise vascular disorders, but rather involves tortuous secretory glands (normal) filled with retained blood but were unable to explain how blood reaches the lumen of the gland, although they were sure that it contains red cells [3]. Vascular dystrophy has been noticed more commonly in women undergoing hysteroscopy during the luteal phase of the menstrual cycle and in patients receiving progestogen therapy. Our patient was receiving progestogens for the management of her heavy bleeding. We did literature search for its association with previous cesarean, endometriosis or adenomyosis as was found in our patient. Isthmocele as a uterine defect allows collection of blood and may result in vascular dystrophy but no data were found as only 10 cases are reported so far.

So we conclude that some findings are very atypical on hysteroscopy and must be diagnosed and reported so that clinicians become aware about existence of this benign condition which does not require any intervention.

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Declarations

Conflict of interest The authors declare that they have no conflict of interest and no funding was involved.

Ethical Approval The ethical clearance was taken from institution committee.

Informed Consent Taken.

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